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VIRUS DISEASES IN PREGNANCY AND CONGENITAL DEFECTS

BY

A. BRADFORD HILL, RICHARD DOLL, T. McL. GALLOWAY, AND J. P. W. HUGHES

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To determine the probability that an illness of the mother during her pregnancy will give rise to a congenital malformation in the infant, essentially demands a prospective method of inquiry. In other words, the attack of illness in the pregnant woman must first be observed and medically diagnosed, and the condition of the child born to her must subsequently be noted. The retrospective approach, *i.e.* noting first the congenital deformity in the new-born child and then obtaining the history of the mother during pregnancy, inevitably omits the normal children whose mothers were affected, and is thus bound to give a highly exaggerated picture of the risks involved. The prospective inquiry is, however, extremely difficult to carry out on an adequate scale. The attack rates from infectious diseases are relatively low in adult life and very large numbers of women must be observed. It is specially important, too, that the occurrence of disease in the mother be recognized and recorded at a time when she may not even know that she is pregnant, *i.e.* during the first month of pregnancy.

One way of securing such records in Great Britain has been described in an earlier paper (Bradford Hill and Galloway, 1949). The data there used were automatically collected by the approved societies operating up to July 5, 1948, under the National Health Insurance Acts. Under these Acts, if an employed and insured married woman drew benefit for a sickness which caused her to be absent from work, she had to present a sickness certificate giving the dates of her illness and the general practitioner's diagnosis of its cause. If she subsequently gave birth to a child (live or still) she would be entitled to draw maternity benefit and the date of birth would therefore also be recorded by the approved society. Thus the dates of the two events would be available in records automatically maintained. By these means information was collected on 22 cases over the space of some 2 years; they included ten cases of rubella and six of measles. The infants were specially examined not long after birth and one case of congenital heart disease following rubella in the first month of pregnancy was observed. The authors concluded their report with the hope that it might "be possible to extend these observations through the records of the new health and social services" (which came into being on July 5, 1948). This hope has been fulfilled. With the aid of the Ministry of Pensions and National Insurance we have been able to collect similar records for 100 women reported to have suffered from rubella, measles, mumps, or chicken pox during, or shortly before, pregnancy. A further seven cases were brought to our notice privately but prospectively (*i.e.* before the birth of the child). It is with this total of 129 "double-events" that the present paper is concerned.

NOTIFICATION OF CASES

The identification of relevant cases followed the system described above. In local offices of the Ministry of Pensions and National Insurance a special note was made on the file of any married woman who returned a sickness certificate signed, as required, by a medical practitioner and bearing a diagnosis of rubella (German measles), morbilli (measles), varicella (chicken pox), or epidemic parotitis (mumps). A form was set up showing the identification particulars of the insured woman and the nature and dates of her illness. If the same woman claimed maternity benefit for the birth of a child within 12 months of the end-date of that illness, then the date of confinement and the name of the doctor or midwife in attendance were added to the form, and notification of the "double event" was made to the Ministry.

In setting up this notification system, steps were specially taken to avoid any breach of medical

confidence. As each case was notified to the Ministry of Pensions and National Insurance a letter from the Chief Medical Officer to the Ministry (or his deputy) was sent to the doctor who had signed the original sickness certificate. This letter enclosed a copy of the completed form relating to his patient and told him that the case was of interest in an inquiry into congenital defects being made by one of us (B. H.) from whom he would be hearing. At the same time the Ministry sent to the Statistical Research Unit similar particulars of the case—dates of illness, etc.—and including the certifying doctor's name but omitting that of the patient.

On receipt of these particulars we immediately communicated with the doctor asking for his help and for permission to visit the mother and baby, known by name to him but not to us. Very few difficulties arose and the mothers were seen and the babies examined by one of us in nearly all instances.

The scheme was operated in London and the home counties from November, 1949, and in a group of midland and north-midland counties* from January, 1951, until the end of March, 1953.

It will be noted that the method of inquiry is limited to the observation of stillbirths and to the condition of liveborn infants; it automatically excludes miscarriages and abortions. If virus diseases in pregnancy contribute to early foetal loss this inquiry cannot reveal it.

DATA

In analysing the data we have calculated the stage of the pregnancy from the first day of the last preceding menstrual period. This is open to criticism in that the first 2 weeks of "pregnancy" may consequently represent a period which actually preceded conception. We have, however, followed this technique, because it is the normal obstetric procedure, which most other investigators have followed, and furthermore, because several instances have been recorded in which rubella manifested itself in the mother during the 2 weeks following the menstrual period and the infant showed the characteristic rubella defects. On the other hand, no instances have been recorded in which the infection appeared before the last menstrual period. In those cases which followed infection in the first 2 weeks, the infection may have persisted until after conception or the wrong date may have been given for the last menstrual period. In the present series, we determined the date of onset of the last menstrual period whenever possible according to the mother's statement. In 44 cases (37 per cent.), however, no such statement was recorded, and, in these, we calculated the date by subtracting 40 weeks from the date of delivery.

Our method of inquiry was based, for administrative convenience, upon a date of delivery that lay within 12 months of the end of the specified illness. It thus automatically produced a number of cases of disease that must have preceded conception by any time up to 3 months. It is, however, of interest to see whether there is any evidence of congenital defects related to an illness *shortly* preceding conception and we have, therefore, examined such babies and included them in the study.

Of the total 129 cases notified to us we subsequently omitted ten from the analysis. In four cases the diagnosis of the illness that occurred during pregnancy was not substantiated when we visited the mother and certifying practitioner (three cases of "measles" in the series reported by Bradford Hill and Galloway and one of "mumps" in the later series). Three cases of measles had occurred too long before conception to be of interest, and one of mumps occurred too close to delivery (4 days). These eight exclusions are obviously called for and are of no importance, but two other cases were lost for different reasons. In one case of chicken pox in the fourth week, the mother refused to allow the child to be examined and no report could be obtained from the family doctor. In one case of mumps in the 11th week, no early examination of the infant was made and the case was subsequently lost to sight.†

The remaining 119 cases are included in the analyses that follow. They concern 44 cases of rubella (6 preceding and 38 during pregnancy), 35 cases of mumps (6 preceding and 29 during pregnancy), 30 cases of chicken pox (6 preceding and 24 during pregnancy), and 10 cases of measles (4 preceding and 6 during pregnancy). One of the women who had chicken pox gave birth to twins so that altogether 120 babies were available for study of whom 110 were personally examined by one of us shortly after birth. In the remaining ten we had information from a hospital or private doctor. Further information was obtained after the child had reached at least 3 years of age. In most cases we again made personal contact, usually with a special examination, and in the remainder we had reports from private doctors, except in two cases in which the family had emigrated and we had to rely upon a report of normality from the mother.

* Staffordshire, Warwickshire, Derbyshire, Leicestershire, Northamptonshire, and Nottinghamshire.

†This child (birth weight 10lb. 4 oz.; 4·7 kg.) was traced after this paper had gone to press; at age 4½ years it had no physical defects.

Results

RUBELLA.—The observations on rubella are set out in Table I and in the following brief case histories:

(1) Start of last menstrual period (mother's statement) 13.4.52, (calculated date) 13.3.52; onset of rubella 24.4.52; delivery 18.12.52. Birth weight 4 lb. 9 oz. (2·1 kg.). Bilateral cataract, harsh systolic murmur all over praecordium. Hospital report "murmur suggestive of ventricular septal defect". Died aged 3 months; certified cause congenital heart disease; no autopsy.

(2) Start of last menstrual period (mother's statement) 9.4.52, (calculated date) 8.4.52; onset of rubella 29.4.52; delivery 13.1.53. Birth weight 6 lb. 2 oz. (2·8 kg.). Bilateral cataract, microphthalmos.

(3) Start of last menstrual period (calculated date) 19.2.47; onset of rubella 17.3.47; delivery 26.11.47. Birth weight 5 lb. 8 oz. (2·5 kg.). Patent interventricular septum, severe bilateral deafness. Deafness not detected at first examination (Hill and Galloway, Case 1).

(4) Start of last menstrual period (calculated date) 7.5.47; onset of rubella 13.6.47; delivery 11.2.48. Birth weight not known. Died aged 5 weeks; certified cause pneumonia. Not examined personally (Hill and Galloway, Case 2).

(5) Start of last menstrual period (mother's statement) mid-January, 1947, (calculated date) 23.1.47; onset of rubella 15.4.47; delivery 30.10.47. Birth weight 6 lb. 7 oz. (2·9 kg.). Partial bilateral deafness, requiring education in a special school for deaf children. Deafness not detected at first examination (Hill and Galloway, Case 5). A test on the mother 4 years after the child's birth suggested that she might also have had a subclinical infection with toxoplasma.

(6) Start of last menstrual period (mother's statement) 31.1.52, (calculated date) 25.1.52; onset of rubella 5.5.52; delivery 1.11.52. Birth weight 5 lb. 6 oz. (2·4 kg.). Mongol.

(7) Start of last menstrual period (mother's statement) 13.12.51, (calculated date) 29.12.51; onset of rubella 28.3.52; delivery 5.10.52. Stillbirth, weight 4 lb. 4 oz. (1·9 kg.). No specific cause found at autopsy. The mother also had an attack of herpes zoster during the pregnancy, starting on 17.3.52.

(8) Start of last menstrual period (calculated date) 5.2.46; onset of rubella 1.7.46; delivery 12.11.46. Birth weight not known. Died aged one week; certified cause pneumonia (Hill and Galloway, Case 8).

TABLE I

OBSERVATIONS ON RUBELLA (GERMAN MEASLES)

Stage at which Rubella Commenced		No. of Cases	No. of Babies Still-born or who Died before Examination	No. of Live-born Babies in whom Abnormalities were Detected	No. of Babies Born weighing 5½ lb. or Less
Before last menstrual period		6	0	0	0
Week of Pregnancy	1st	2	0	0	0
	2nd	1	0	1	1
	3rd	2	0	1	0
	4th	2	0	1	1
	5th	2	0	0	0*
	6th	2	1	0	0*
	7th	1	0	0	1
	8th	1	0	0	0
	9th	0	—	—	—
	10th	0	—	—	—
	11th	2	0	0	0
	12th	0	—	—	—
	13th	3	0	1	0
	14th	3	0	1	1
	15th	0	—	—	—
	16th	1	1	0	1
	17th	2	0	0	0
	18th	1	0	0	0
	19th	0	—	—	—
	20th	0	—	—	—
	21st	3	1	0	0*
	22nd	0	—	—	—
	23rd	3	0	0	0
	24th	0	—	—	—
	25th	0	—	—	—
	26th	2	0	0	0
	27th or Later	5	0	0	0
Total	..	44	3	5	5

* Birth weight not available for one child.

It will be seen that, of the seven infants whose mothers had rubella in the first 4 weeks of pregnancy, three had major and characteristic abnormalities. There were eleven cases of rubella occurring in the next 9 weeks, *i.e.* up to the end of the first trimester, and amongst these was one baby who died aged 5 weeks unexamined and one child who was deaf. As regards the first trimester, therefore, there were certainly four and possibly five affected babies in a total of eighteen. Of the fifteen infants whose mothers were infected in the second trimester, one was stillborn, one was a mongol, and one died at the age of one week, unexamined. A fourth child was found at the second examination, at the age of 4¼ years, to have a moderately loud systolic murmur at the left sternal edge, varying in intensity with the position of the child; in the absence of any other signs, this has been regarded as physiological. The mother had had rubella in the 24th week of her pregnancy and the child's birth weight was 7 lb. 10 oz. (3·4 kg.). Of the five infants whose mothers were infected in the third trimester, none showed any defect. This was also the case with the six infants whose mothers had had rubella before the last menstrual period.

Of the eight children known to be defective or who died before examination, the birth weight was available in six; four weighed 5 lb. 8 oz. (2·5 kg.) or under, and none weighed as much as 7 lb. (3·2 kg.).

Of the 36 normal children, 33 were known to be alive and well when aged 3 years or more; one was certified as having died at 5 months from "asphyxia due to overlaying" (maternal rubella one month before the last menstrual period, birth weight 8 lb. (3·6 kg.)) and two were not traced after the initial examination. The birth weight was available in 35; one baby weighed 4 lb. 9 oz. (1·8 kg.), eight weighed over 5½ and less than 7 lb. (2·5 to 3·2 kg.), and 26 weighed 7 lb. (3·2 kg.) or over.

MUMPS.—The observations on mumps are set out in Table II and in the accompanying footnotes. One congenital defect (bilateral talipes equino-varus) was noted in the 29 instances in which the disease occurred during pregnancy. One child died of "white asphyxia" at the age of 3 days and the autopsy revealed no other specific defect. All the six infants whose mothers were infected before the last preceding menstrual period survived and no defects were observed.

Of the 33 normal children, 31 were known to be alive and well when aged 3 years or more, one had suffered from Still's disease and had died when aged 3¼ years, and one was not traced after the initial examination. At birth, one of the normal children weighed 5 lb. 1 oz. (2·3 kg.), five weighed over 5½ and less than 7 lb. (2·5 to 3·2 kg.), and 26 weighed 7 lb. (3·2 kg.) or more; the weight of one child was not known.

TABLE II

OBSERVATIONS ON MUMPS (EPIDEMIC PAROTITIS)

Stage at which Mumps Commenced		No. of Cases	No. of Babies Stillborn or who Died before Examination	No. of Liveborn Babies in whom Abnormalities were Detected	No. of Babies Born weighing 5½ lb. or Less
Before last menstrual period		6	0	0	0
Weeks of Pregnancy	1st– 4th	1	0	0	0
	5th– 8th	10	0	0	0
	9th–12th	5	0	1(*a*)	0
	13th–16th	2	1(*b*)	0	0
	17th–20th	1	0	0	0
	21st–24th	3	0	0	1
	25th or Later	7	0	0	0*
Total		35	1	1	1

* Birthweight not available for one child.

(*a*) Start of last menstrual period (mother's statement and calculated date) 1.8.51; onset of mumps 2.10.51; delivery 8.5.52. Birth weight 7 lb. 12 oz. (3·5 kg.). Bilateral talipes equino-varus.

(*b*) Start of last menstrual period (mother's statement) 21.7.50 (calculated date) 8.8.50; onset of mumps 21.10.50; delivery 15.5.51. Birth weight 6 lb. 11 oz. (3·0 kg.). Died on 3rd day with "white asphyxia"; no abnormalities detected at autopsy.

CHICKEN POX.—The observations on chicken pox are set out in Table III. The only congenital defect noted was an isolated defect of pancreatic secretion, and in this case the mother's infection had occurred 4 weeks before the start of the last menstrual period. Two babies were stillborn. Autopsy in one case showed atelectasis and suprarenal haemorrhage, with no specific congenital abnormality. The second foetus was 5 weeks premature and was macerated and a large clot was found behind the placenta. It may be recalled that another stillbirth followed an attack of herpes zoster in the 14th week of pregnancy, when the mother had also suffered from rubella in the 15th week (Case 7).

Of the 28 normal children, 24 were known to be alive and well when aged 3 years or more, and four were not traced after the initial examination. Birthweights were available for 26 of the children; one weighed 5 lb. 6 oz. (2·4 kg.), 12 weighed over 5½ and less than 7 lb. (2·5 and 3·2 kg.) and 13 weighed 7 lb. (3·2 kg.) or over.

TABLE III

OBSERVATIONS ON CHICKEN-POX (VARICELLA)

Stage at which Chicken-pox Commenced		No. of Cases	No. of Babies Stillborn or who Died before Examination	No. of Liveborn Babies in whom Abnormalities were Detected	No. of Babies Born weighing 5½ lb. or Less
Before last menstrual period		6	0	1(*a*)	0
Weeks of Pregnancy	1st– 4th	1	0	0	0
	5th– 8th	3	0	0	0*
	9th–12th	3	1(*b*)	0	0
	13th–16th	5	0	0	0
	17th–20th	2	1(*c*)	0	1
	21st–24th	3	0	0	0
	25th or Later	8	0	0	1
Total		31†	2	1	2

* Birthweight not available for two children.

† Including one pair of twins (chicken-pox in the 5th week); excluding one normal child whose mother had chicken-pox in the 17th week following rubella in the 5th week, and one stillborn child whose mother had herpes zoster in the 14th week and rubella in the 15th week (Rubella Case 7).

(*a*) Start of last menstrual period (mother's statement and calculated date) 22.6.49; onset of chicken-pox 24.5.49; delivery 29.3.50. Birthweight 7 lb. 11 oz. (3·5 kg.). Isolated defect of pancreatin secretion.

(*b*) Start of last menstrual period (mother's statement) 25.4.50, (calculated date) 29.4.50; onset of chicken-pox 10.7.50; delivery 3.2.51. Birthweight 9 lb. 3 oz. (4·2 kg.). Stillborn; atelectasis and suprarenal haemorrhage found at autopsy; no other abnormality.

(*c*) Start of last menstrual period (mother's statement) 15.11.51, (calculated date) 12.10.51; onset of chicken-pox 24.3.52; delivery 19.7.52. Birthweight 4 lb. 6 oz. (2·0 kg.). Stillborn; macerated foetus with large clot behind the placenta.

MEASLES.—As nearly all children in Great Britain have measles, an attack is relatively rare in adult life, and Table IV gives data for only ten cases. In

four of these, the attack of measles began before the menstrual period which preceded the pregnancy. Two congenital defects were observed—a right talipes equino-varus following measles that occurred one day before the start of the last menstrual period, and a cerebral diplegia following measles in about the 25th week of pregnancy. In the latter case, the mother had also shown signs of toxaemia one month before delivery. Of the eight normal children, six were known to be alive and well when aged 3 years or more, and two were not traced after the initial examination. Birthweights were available for six of the children; one weighed 4 lb. 5 oz. (2·0 kg.) and the others all weighed 7 lb. (3·2 kg.) or over.

TABLE IV

OBSERVATIONS ON MEASLES (MORBILLI)

Stage at which Measles Commenced		No. of Cases	No. of Babies Stillborn or who Died before Examination	No. of Liveborn Babies in whom Abnormalities were Detected	No. of Babies Born weighing 5½ lb. or Less
Before last menstrual period		4	0	1(*a*)	0
Weeks of Pregnancy	1st– 4th	1	0	0	0
	5th– 8th	0	—	—	—
	9th–12th	0	—	—	—
	13th–16th	1	0	0	0*
	17th–20th	2	0	0	0
	21st–24th	0	—	—	—
	25th or Later	2	0	1(*b*)	0*
Total		10	0	2	0

* Birthweight not available for one child.

(*a*) Start of last menstrual period (mother's statement) 15.4.49, (calculated date) 26.4.49; onset of measles 14.4.49; delivery 31.1.50. Birthweight 6 lb. 3 oz. (2·8 kg.). Right talipes equino-varus.

(*b*) Start of last menstrual period (calculated date) 28.9.46; onset of measles 27.3.47; delivery 5.7.47. Birthweight 7 lb. 9 oz. (3·4 kg.). Cerebral diplegia. The mother also had mild toxaemia one month before delivery.

DISCUSSION

The present investigation is clearly not extensive enough to provide an accurate estimate of the risk of malformation following an attack of rubella at different stages during pregnancy. Reports have, however, been published of several other investigations in which a prospective method of inquiry has been strictly applied, and the results of these studies may be combined. The combined results of four such studies, reported by Brown and Nathan (1954), Brawner (1955), Pitt (1957), and the present authors, are shown in Table V. They comprise—with one exception—all the studies available to us which satisfy the following criteria:

(1) the occurrence of the attack of rubella was in every case recorded *before* the child was born;

(2) the children born to nearly all the affected mothers were subsequently examined;

(3) the data are published in sufficient detail for a separate estimate to be made of the risk for each *month* of pregnancy.

One other inquiry which satisfies these criteria was reported by Lamy and Seror (1956). Their results, however, are so different that it is preferable to consider them separately.

Table V shows that the risk of abnormality may be as high as 50 per cent. when the infection occurs in the first month of "pregnancy" (that is, in the 4 weeks following the start of the last preceding menstrual period). Subsequently, as the length of time between the last menstrual period and the appearance of the disease increases, the risk of abnormality steadily decreases. Of the nineteen mothers showing the disease in or after the 25th week, none gave birth to a defective child. Not all the defects included, however, can be attributed with certainty to the maternal disease. Of the eleven infants with major defects with whom the maternal disease had occurred within the first 2 months of pregnancy, all showed a congenital heart disease, cataract, or deafness. It may be presumed, therefore, that all these defects were the result of the infection. Of the three infants with defects whose mothers were reported to have been ill in the third month, two died of a congenital heart lesion and one had a hare lip and cleft palate. Of the three infants whose mothers were reported to have been ill in the fourth, fifth, and sixth months, one showed bilateral deafness (the mother's illness occurred in the 13th week of pregnancy), and the other two were respectively a mongol (maternal illness, 14th week) and an anencephalic (maternal illness, 21st week). It is possible that the last two defects were not related to the preceding rubella, and that therefore no defects were due to the maternal illness when it occurred after the 13th week of pregnancy (some fifty cases in all). On the other hand, a number of lesions attributable to the disease may have been omitted from the various inquiries represented in Table V. One foetus was stillborn and macerated following rubella in the 6th week, and another was stillborn, without any specific lesion being found at autopsy, following rubella in the 15th week. Two children died within 6 weeks of birth without being specially examined (following rubella in the 6th and 21st week), and one child was thought to have a "possible mild grade of pulmonary stenosis" following rubella in the 14th week (Pitt, 1957). Moreover, some of the children were examined on only one occasion and at a relatively early age

(1 year or less) and cases of deafness may thus have been missed.

TABLE V

RISK OF DEFECT IN THE INFANT FOLLOWING MATERNAL RUBELLA DURING PREGNANCY

(Summation of data from four series)

Stage of Pregnancy at which Rubella Commenced		Number of Cases	Infants with Major Defects	
			Number	Per cent.
Weeks of Pregnancy	1st– 4th	12	6	50
	5th– 8th	20	5	25
	9th–12th	18	3	17
	13th–16th	18	2	11
	17th–24th	17	1	6
	25th or Later	19	0	0
Total		104*	17	—

* Excluding two cases which terminated in abortion (rubella in the 5th and 12th weeks respectively) and three cases in which the children could not be traced. One pair of fraternal twins (rubella in the 21st week) is included as two cases.

The results of the inquiry of Lamy and Seror (1956), which were excluded from Table V, are summarized in Table VI. In this study, a questionnaire was sent to over 100,000 insured women who stated they were pregnant between 1.3.53 and 31.3.54. The questionnaires were sent out in the fifth month of the pregnancies. Nearly half the women replied: fifty stated that they had had rubella in the first 4 months of the pregnancy and 48 of these could be followed up. The results suggest that the risk to the foetus may be substantially greater than was observed in the four studies summarized in Table V.

TABLE VI

OBSERVATIONS OF LAMY AND SEROR (1956) ON EFFECTS OF MATERNAL RUBELLA DURING PREGNANCY

Stage of Pregnancy at which Rubella Commenced		Number of Cases	Number ending in Abortion	Infants with Major Defects	
				Number	No. as per cent. of Cases not ending in Abortion
Weeks of Pregnancy	1st– 4th	7	0	6	86
	5th– 8th	21	5	13	81
	9th–12th	12	0	3	25
	13th–20th	8	1	3	43
Total		48	6	25*	—

* One other child was stillborn (rubella in the 8th week) and one died at the age of 15 days, cause unknown (rubella in the 11th week).

Other studies, however, have suggested that the risk may be lower. Greenberg, Pelliteri, and Barton (1957), for example, have summarized the data from seven series not included in Table V. Among 96 infants born to mothers affected by rubella during the first trimester of pregnancy, six were stillborn (6·3 per cent.) and only seven showed any major congenital defect (7·3 per cent.). In most of the series it is not possible to distinguish between the risks in the various months of the first trimester, and, since rubella in the early months has been regarded as an indication for abortion, it is possible that the data are disproportionately weighted with cases occurring towards the end of that trimester. This, however, cannot account for the low incidence of defects reported by Lundström (1952). An epidemic of rubella occurred in Sweden in the spring of 1951 and the staffs of all the Swedish maternity hospitals were requested to question all women who were delivered in them or who were treated for a spontaneous abortion during the following year. Altogether 1,067 women reported that they had had rubella during pregnancy. The proportion of stillbirths, neonatal deaths, and congenital abnormalities rose from 10 per cent. when the disease was contracted in the first month, to 14 per cent. for the second month; it fell again to 10 per cent. for the third month and finally to about 3 per cent. for the last 4 months of pregnancy. The corresponding figure for a control group of 2,452 children born in the same hospital at the same period was approximately 4 per cent.

The great differences in the results obtained by different groups of workers may be explained, in part, by the operations of chance and, in part, by differences in the methods used to investigate the problems and in the presentation of the results. It seems unlikely, however, that all the differences can be thus explained, and it is possible that the effect of a maternal attack of rubella may vary in space and time.

The results obtained in the present study from the follow-up of mothers who were infected during their pregnancy with mumps, chicken pox, or measles, provide no clear evidence of any deleterious effect of these diseases on the foetus. The only possible exception was chicken pox. Half the mothers who suffered from this disease gave birth to children weighing less than 7 lb., compared with 22 per cent. of the normal infants born to mothers who had one of the other diseases. The mean birth weights were not, however, significantly different (7·09 lb. for chicken pox and 7·43 for the other diseases: "t"=1·48; *P* greater than 0·1). Other workers (for example, Grönvall and Selander, 1948) failed to observe any characteristic effect from these infections and it seems very unlikely that they carry any important risk to the foetus.

SUMMARY

By a special recording system the occurrence of certain infectious diseases was noted in women whose illness, medically observed and diagnosed, fell during, or shortly before, a pregnancy. Information was subsequently sought regarding the infants born to them (live or still but excluding abortions or miscarriages), shortly after birth and again at an age of not less than 3 years. Data were thus acquired on 44 cases of German measles (rubella), 35 of mumps (epidemic parotitis), 30 of chicken pox (varicella), and 10 of measles (morbilli). There was no evidence that mumps or measles had had any deleterious effect upon the foetus. With chicken pox there was also no evidence of the production of congenital defects, but the proportion of liveborn children with low birth weights was relatively high (though the difference was not statistically significant). With rubella occurring early in pregnancy, the well-known congenital defects of heart, vision, and hearing were observed.

By the addition of these present cases to similar published records, some estimates are reached of the risks involved with rubella in different stages of pregnancy. An incidence of six affected children out of twelve with rubella in the mother in the first month of pregnancy (50 per cent.) declines to five out of twenty for the second month (25 per cent.) and to three out of eighteen for the third month (17 per cent.). With 54 women in whom the attack of rubella occurred in the second and third trimesters, 51 infants were unaffected, and only one of the three affected cases seemed likely to be attributable to the preceding rubella.

Other published data have shown both considerably higher and considerably lower incidence rates of congenital defects following rubella in early pregnancy. It may be therefore that the risk varies in space or time.

We are indebted to the Ministry of Pensions and National Insurance for putting in operation the system by which we were notified of the majority of the cases, and in particular to Sir Arthur Massey, the Chief Medical Officer, for much help and advice; we are also indebted to the doctors who notified us of the seven cases which were brought to our attention privately, to Dr. R. Burtles who, while working in the Statistical Research Unit, carried out the second examination on a number of the children, to the practitioners and specialists who examined some cases on our behalf, and to Miss Keena Jones, of the Statistical Research Unit, who traced the whereabouts of many of the children and who interviewed some of the elder children and their mothers.

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ENVIRONMENTAL INFLUENCES RELATED TO THE AETIOLOGY OF CONGENITAL DISLOCATION OF THE HIP

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Congenital dislocation of the hip has been recognized for many centuries and has given rise to a literature so extensive that a comprehensive review cannot be attempted here. The condition was described and named by Hippocrates, who suggested uterine pressure and birth trauma as possible causes.

Paré published a more detailed description of the defect and recognized its occasional hereditary nature. Extensive clinical and pathological studies were made during the 18th and 19th centuries but failed to suggest any more precise explanation than that of a diathesis, manifestations of which depended on various environmental influences. The only evidence of environmental hazard related to breech presentation, which was believed to predispose to dislocation either by foetal deformation or obstetric trauma. An excellent review by Sainton (1893) covers the early history.

Vogel (1905) made the first detailed study of the influence of heredity and of the uterine environment, finding that an affected relative could be remembered in about a third of the families studied, and that breech presentation and other obstetric difficulties were unduly common in the births of affected children. Le Damany (1912, 1914) made a major advance in distinguishing cases developing as a consequence of gross abnormalities, such as spina bifida, from the majority which he regarded as merely one end of a continuously variable distribution of hip stability. In his elaboration of Darwin's view of malformations as extreme varieties, he may claim precedence in applying the concepts of quasi-continuous variation and polymorphism to man. His views on congenital dislocation of the hip are strictly analogous to contemporary views of mental deficiency, which may either result from metabolic abnormality or local disease, or be merely the lower end of a continuous distribution.

Unfortunately, these two papers appeared at a time when human genetics was passing through a phase of rapid change. Effort was concentrated in attempting to apply Mendel's concepts to the inheritance of human characteristics and diseases, and workers tended to think in terms of single gene substitutions. Environmental influences were largely ignored and genetical hypotheses of increasing complexity were formulated in an attempt to satisfy observational data. Isigkeit (1928) for example, suggested that congenital dislocation of the hip was determined by two sex-linked dominant genes (one of which was presumed to be semi-lethal when recessive) and a pair of recessive autosomal genes. Faber (1937) concluded, from radiographic measurements of relatives of affected persons, that a dysplasia of the hip, defined in terms of these measurements, is inherited as a Mendelian dominant of incomplete penetrance. This view, strongly supported by Hart (1947), received wide acceptance and is considered by some to be so well established as to form the basis for eugenic counselling and even for the sterilization of so-called latent carriers. Faber's interpretation of his data, however, is open to doubt because the measurements show no evidence of bimodality. In fact, the data appear to show little more than that in about half the cases the measurements were below the average of the whole series. The limitations of the interpretation may be illustrated by considering a hypothetical disease predisposed to by tallness. If measurements of relatives of a few affected persons were made, it would be possible to define tallness in such a way that it would appear to be transmitted as a Mendelian dominant of incomplete penetrance.

A careful twin study by Idelberger (1951) may help to re-orientate opinion in a more promising direction. An examination of 138 living twin pairs showed that concordance occurred in less than half the monozygous pairs, and that among dizygous pairs the twin of an affected member was no more likely to have the defect than any other sibling of the

same sex. Although some of Idelberger's conclusions are not acceptable, his observations are invaluable and seem to be consistent with the view of earlier workers that a number of factors, both genetical and environmental, must be implicated. It seems doubtful whether the hereditary background will be understood until more is known of environmental influences, and the present work was planned to explore these factors.

Material

An attempt was made to assemble a comprehensive series of cases of congenital dislocation of the hip occurring in children born in the years 1942–52. This period was chosen because an estimate of the maternal age and parity distribution of all Birmingham births for these years was already available from a previous investigation (Smith and Record, 1955). Inquiry at all hospitals in Birmingham indicated that treatment for congenital dislocation of the hip was undertaken at only the Children's Hospital and the Royal Orthopaedic Hospital. The diagnostic indexes of these hospitals provided the names and addresses of 202 patients whose dates of birth came within the specified period (Table I). Sixteen lived more than 12 miles away and were excluded from the survey. An attempt was made to visit the homes of the remaining 186 patients. In nineteen cases the family had moved and could not be traced. The rest of the series (167 cases) were visited and the mothers were interviewed. These cases provide the data on which most of the analysis is based, but the whole series of 186 propositi has been used to examine seasonal variation in incidence, the date of birth being known for all the children. For those parts of the analysis which required knowledge of the related population, attention was restricted to propositi whose parents resided in Birmingham when the affected children were born; patients whose parents lived outside the city were excluded since they came from an area with no precise boundaries. The estimate of incidence, therefore, is based on the 148 Birmingham cases, and the examination of the influence of maternal age and parity is restricted to the 136 who were visited.

Table I

CASES OF CONGENITAL DISLOCATION OF THE HIP BORN IN THE PERIOD 1942–52

Number of Affected	Domicile at Time of Birth		Total
	Birmingham	Elsewhere	
In Hospital Records ..	148	54	202
Excluded from Survey* ..	—	16	16
In Survey	148	38	186
Untraced	12	7	19
Whose Parent was Interviewed	136	31	167

* Because domicile was more than 12 miles from Birmingham.

Results

Incidence

The 148 cases among Birmingham children born in the years 1942–52 may be related to the total number of live births (226,038) computed from data published annually in the Registrar General's Statistical Reviews. This gives an incidence of 0·65 per 1,000 live births (0·2 and 1·1 per 1,000 for males and females respectively). This is lower than most published estimates, but it is well known that incidence shows very marked regional variations. Among the Lapps, incidence is said to be about 4 per cent. (Getz, 1955), and considerably higher values have been reported in some Lapp villages (Allison, 1956), and in Red Indian settlements (Steinberg, 1957). The condition is rare among Negroes and almost unknown among the Sudanese. It is possible that most recorded estimates come from areas of high prevalence where the condition is more likely to be studied because of its more obvious social and economic importance.

It may be advisable, however, to consider the reliability of the present estimate. Ascertainment may have been incomplete for the following reasons:

(1) Some affected infants may have died before the abnormality was detected. There is undoubtedly a relationship between congenital dislocation of the hip and several conditions such as breech presentation and low birth weight which are associated with raised perinatal mortality.

(2) Some children may have moved outside the Birmingham area before the defect was recognized. It seems likely that this was rare. Among the 167 affected in the series who were traced there was only one whose parents had lived outside Birmingham when the child was born and had later moved into the city. It is reasonable to suppose, therefore, that not more than one or two cases were lost by emigration, since during the period of the survey more people were coming into the city to live than were leaving it.

(3) At the time when the present investigation was begun the condition may not have been diagnosed in some children born in the later years of the period. We believe that such losses are very small since the survey was not started until the youngest children had attained the age of 3 years. 85 per cent. of the

cases in the series had been diagnosed by the age of 2 years, and only 3 per cent. were diagnosed after the third birthday.

(4) Some cases in older children may not have been diagnosed, or the diagnosis may have been made too late for treatment to be attempted. We have no knowledge of the number of these cases, but in view of the efficiency of the Child Welfare and School Medical Services in Birmingham it seems certain that it is small.

It seems reasonable to conclude that the figure of 0·65 per 1,000 births for the incidence of congenital dislocation of the hip in Birmingham may be slightly below the true value, but is probably acceptable as an estimate of incidence among children who survived infancy.

Characteristics of Propositi

(*a*) *Sex Ratio.*—The complete series consisted of 27 males and 159 females. The proportion of males (14·5 per cent.) is in close agreement with the sex ratio (14·9 per cent.) estimated by Poli (1937) on 37,503 cases reported in the literature. The higher incidence in females was considered by le Damany (1912) to be due to sex differences in the structure of the pelvic bones and particularly in the inclination of the acetabula. This view was supported by Dega (1933), who found sex differences in the shape of the foetal pelvis, even as early as the third month.

(*b*) *Side Affected.*—In two cases details of the affected side were not available. Among the remaining 184, the condition was bilateral in 47 (25·5 per cent.) and involved the left hip only in 101 and the right hip only in 36. The number of affected hips in the series was, therefore, 231, of which 148 (64·1 per cent.) were left-sided. Nearly all published series show a higher incidence on the left side than on the right, but the discrepancy is not usually as marked as in the present series. There appears to be little difference between males and females in the relative frequency of the double lesion (Table II).

Table II

AFFECTED SIDE ACCORDING TO SEX

Affected Side	Males		Females*		Total	
	No.	Per cent.	No.	Per cent.	No.	Per cent.
Left only ..	15	55·6	86	54·8	101	54·9
Right only ..	4	14·8	32	20·4	36	19·6
Both ..	8	29·6	39	24·8	47	25·5
Total	27	100	157	100	184	100

* Two cases for whom the affected side was not stated have been excluded.

There is even less difference between the sexes in the frequency of involvement of the left hip relative to the right; the 27 male propositi had 35 affected hips, of which 23 (66 per cent.) were left-sided; the 159 female propositi had 196 dislocations, of which 125 (64 per cent.) were left-sided.

(*c*) *Associated Defects.*—Details of associated abnormalities were obtained during the home visit and are based, therefore, on the 167 propositi who were traced. Defects, details of which are given in Table III, were present in 26 (16 per cent.). Although no strictly comparable rate is available for the general population, there seems little doubt that this incidence is very high, and is the more remarkable when it is recalled that the series consisted of children who survived infancy and would, therefore, be unlikely to exhibit the more serious malformations. Grundy and Lewis-Faning (1957), in an extensive survey of infants born in 1952 in fifteen areas of England and Wales, found a malformation rate among live births of 2·9 per cent., and one-quarter of these infants died before the first birthday. There are many reports in the literature of the frequent association of congenital dislocation of the hip with other deformities, but an incidence as high as 16 per cent. is rare. Another interesting feature is that three of the 26 propositi with associated defects were twins. The defects were hare-lip and cleft palate, torticollis, and diaphragmatic hernia. Since the whole series contained only four twins, the risk of associated defects appears to be much greater for twins than for single births, but this view is not supported by the observations of Idelberger (1951).

Table III

ASSOCIATED DEFECTS OCCURRING AMONG 167 PROPOSITI

Type of Defect	Diagnosis	No.	Per cent.
Nervous	Spina Bifida and Hydrocephalus	1	4·2
	Spastic Paralysis of Hand	2	
	Spastic Paralysis and Mental Defect	2	
	Mental Defect	1	
	Amyotonia	1	
Musculo-Skeletal	Diaphragmatic Hernia	1	10·2
	Other Herniae	3	
	Torticollis	2	
	Dislocation of Patellae	1	
	Genu Valgum	1	
	Talipes	8	
	Pes Planus	1	
Other	Hare-lip and Cleft Palate	1	1·2
	Hypospadias	1	

(*d*) *Birth Weight.*—The weights of the propositi at birth, which were obtained from the mother, were available for 161 of the 167 cases visited. The

distribution of weight was compared with that derived from a survey of all births in Birmingham in 1947 (assembled by Gibson and McKeown, 1950), from which all stillbirths and infant deaths have been excluded. Because of the variation of birth weight with sex and birth rank it was necessary to standardize the controls to make them comparable in respect of these two variables with the series of propositi. When the percentage distribution of affected children is compared with that of the standardized controls, there is an excess of propositi weighing less than 7 lb. and a deficiency of those weighing 9 lb. and over (Fig. 1). The difference between the two distributions is also shown by a comparison of the means. The mean birth weight of affected children was 6·81 lb. and that of the standardized controls 7·36 lb. (difference 0·55 ± 0·09).

This difference, although apparently slight, implies a very considerable increase in risk to small babies. It may be shown that, if birth weight were normally distributed and the affected and the controls had the same standard deviation of about 1 lb., then a difference in birth weight of d lb. would make a difference in relative liability of about $e^{0 \cdot 88d}$; that is, there would be a doubling in incidence associated with each 1·3 lb. deficiency in birth weight. This does not, of course, imply that influencing birth weight would necessarily influence liability to dislocation, as babies prone to dislocation may form a distinct sup-group.

It may be objected that a comparison between the two series is not valid because the weights of propositi were obtained by interrogation of the mother, and the control series was based on observed weights. We have evidence from another inquiry that the great majority of mothers accurately remember the birth weights of their children. The weights of all infants born in a neighbouring town (Smethwick) during one year was recorded. Two years later, the mothers were visited and asked what the birth weight was. 907 mothers were interviewed; a correct statement was obtained from 536 (59 per cent.) and a further 221 (24 per cent.) were not more than 4 oz. in error. Errors greater than a pound were recorded for only eighteen (2 per cent.). The errors of those who understated the weight (193) were very nearly cancelled by those who overstated it (178), and the net effect on the mean weight of the whole series was an underestimation of only 0·005 lb.

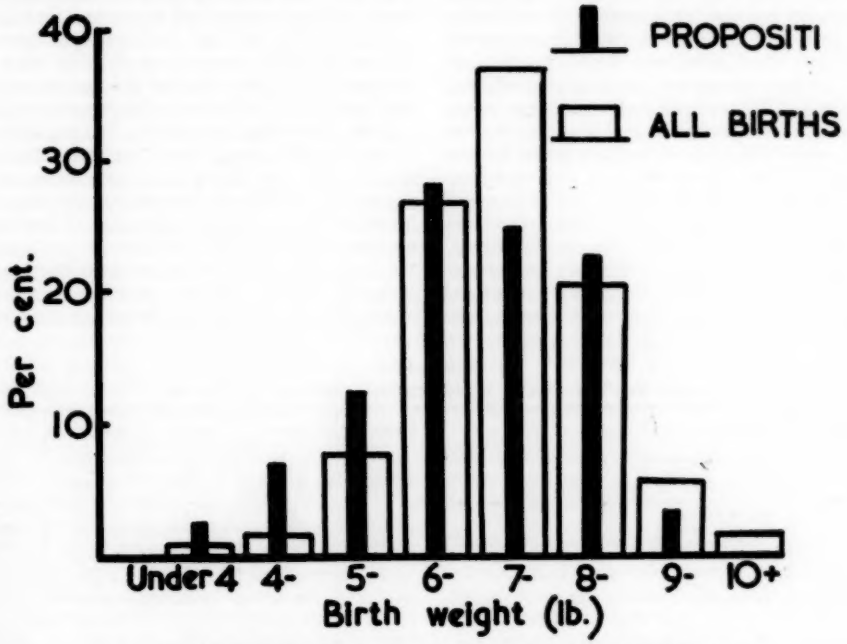

FIG. 1.—Distribution according to birth weight compared with that of all Birmingham births in 1947 (first-year survivors) of the same sex and parity distribution.

There seems to be little doubt, therefore, that infants with congenital dislocation of the hip were lighter than average at birth. This result leads one to inquire whether they were born earlier than usual. Our data are hardly adequate to answer this question with certainty. Mothers were interrogated about the duration of the pregnancy, but could not, of course, be expected to remember the date of the preceding menstrual period, and rather approximate answers such as "full time" or "about the expected date" were often received. Of 165 women who attempted a reply, 140 (85 per cent.) gave histories of pregnancies lasting at least 39 weeks. The corresponding figure derived from the 1947 Birmingham survey (after excluding stillbirths and infant deaths) was 82 per cent., but this was a more reliable estimate, since durations of pregnancy were calculated from the last menstrual period. This rather inadequate evidence suggests that there is nothing unusual about the gestation periods of propositi. This conclusion is not consistent with results reported by Vogel (1905), who found that 24 per cent. of his cases were born at least a fortnight early.

Association with Maternal Age and Birth Rank

In order to examine the variation in incidence with maternal age and birth rank in circumstances where the use of the Greenwood-Yule method is inappropriate (McKeown and Record, 1956), we require to know the age and birth rank of affected children and of the population of births from which the series was assembled. The analysis which follows is based on the 136 propositi whose parents lived in Birmingham at the time of the birth and were later traced, and a control group of 1,156 births (live and still) sampled from the 231,619 infants born to Birmingham residents during the years 1942–52. When the data are set out in a maternal age and birth rank contingency table (Table IV), the ratio of the number of propositi to the number of controls in each cell can readily be converted into an estimate of incidence by multiplying by the ratio of the total number of controls (1,156) to the total number of births (231,619). (The total incidence thus calculated differs slightly from that given in the previous section because the related population includes stillbirths, and because it ignores the twelve Birmingham cases which were not traced.)

The most prominent feature of the data is the relatively large proportion of affected first-born (50·4 per cent. compared with 34·3 per cent. in the control series). The raised incidence among first-born occurs in all the maternal age groups. The liability of first-born to dislocation appears to be about double that of second and third-born. A primogeniture effect of this magnitude does not appear to have been previously recorded for any malformation.

The influence of maternal age is less marked. Incidence among first born rises with increasing maternal age, but for second and third children there seems to be no association. In the higher birth ranks, numbers in the younger age groups are too small to show a trend, but incidence is high among children born to women aged 35 and over.

It may be asked whether the increased risk among first-born can be explained by the greater incidence among children of low birth weight, since first children generally weigh less at birth than later children. We have explored this problem by re-distributing the 1947 series of Birmingham births to give the same weight distribution (taking account of sex) as the series of propositi. The method is simple, being merely a standardization procedure, but the calculation is rather too lengthy to reproduce here. Its effect was to raise the percentage of first-born in the 1947 series from 40·1 to 42·9, *i.e.* an increase of 2·8, which is small compared with the observed difference of 16·1 (50·4 − 34·3 per cent.). It is concluded that the increased frequency of congenital

Table IV

INCIDENCE ACCORDING TO MATERNAL AGE AND BIRTH RANK

Birth Rank	Maternal Age (yrs)	Under 25	25–29	30–34	35 and Over	All Ages
1	No. of Propositi	30	24	12	2	68
	No. of Controls	200	126	46	25	397
	Incidence per 1,000 Live Births	0·75	0·95	1·30	(0·40)	0·85
2 and 3	No. of Propositi	12	13	14	5	44
	No. of Controls	125	187	159	79	550
	Incidence per 1,000 Live Births	0·48	0·35	0·44	0·32	0·40
4 and Over	No. of Propositi	0	3	3	18	24
	No. of Controls	2	42	68	97	209
	Incidence per 1,000 Live Births	—	0·36	0·22	0·93	0·57
All Birth Ranks	No. of Propositi	42	40	29	25	136
	No. of Controls	327	355	273	201	1,156
	Incidence per 1,000 Live Births	0·64	0·56	0·53	0·62	0·59

dislocation of the hip among first-born cannot be accounted for by low birth weight.

There is no evidence in the Age-Parity Table of any association with social class. Variation from this source would be expected to produce a diagonal stratification of incidence in Table IV, since births in poor circumstances are represented mainly towards the left lower corner and those among the well-to-do towards the right upper corner.

Maternal Fertility

The slight association between incidence and advancing maternal age suggests the possibility of some impairment of fertility of mothers of propositi. Data relating to mothers of controls suitable for an examination of this problem were collected for a previous inquiry (McKeown, MacMahon, and Record, 1951a), but unfortunately are complete only for births before 1948. In order to make the series of affected children comparable with this control group, all propositi born after 1947 have been excluded, leaving 87 mothers for consideration. Date of marriage was recorded for 84 mothers. The mean age at marriage was 22·8 years for these women and 22·5 years for mothers of the controls. The difference seems to be too small to account for any difference in fertility between the two groups.

The interval preceding the conception of propositi was estimated for first-born from the date of marriage, pre-marital conceptions being excluded. For later-born propositi the interval was calculated from the date of the preceding birth. Data relating to 78 mothers of propositi were available and the mean fallow period was 38·5 months. The corresponding figure for the mothers of the controls standardized to the same parity distribution was 31·1 months. A measure of the mean relative fertility of the mothers of affected children can be obtained by calculating the ratio of the reciprocals of the two means. This gives a value of 0·81.

Fallow periods preceding conceptions of children born before propositi were calculated in the same way. Mothers of affected had had 53 earlier conceptions and the mean preceding fallow period was 22·8 months. The fallow period for mothers of controls standardized for parity was 18·5 months. The mean relative fertility of mothers of affected was again 0·81.

It appears, therefore, that women who give birth to children with congenital dislocation of the hip are, on the whole, slightly less fertile than other mothers. This conclusion is consistent with the finding that incidence among children of a given birth rank tends to rise with increasing maternal age. It does not, of course, permit one to decide whether mothers of affected children are older because they are less fertile than other mothers or are less fertile because they are older.

Evidence bearing on this problem may be obtained by investigating fertility after the birth of an affected child. Some caution in interpreting the pattern of subsequent reproduction is necessary because of the possibility that it may be influenced by eugenic considerations, but if one may judge from studies of the more lethal malformations, such as those involving the central nervous system, or of the socially less acceptable defects such as mongolism, it would appear that parents are rarely deterred from further reproduction by the possibility that a later child may also be affected.

Mean fallow period is not an efficient index of subsequent fertility; a more useful measure is the number of children born after the propositus in a given time. The control series used above, based on interrogation of mothers in 1948, is not suitable for this investigation because of the relatively short period between the births of propositi and the home visit. Instead we have used a control group which was assembled for comparison with a series of mongols (Smith and Record, 1955). This group consisted of 139 mothers of children born in the years 1942–52; information on the number of subsequent children born by the end of 1953 was obtained by home visits. It was selected from the original control series of 1,156 births on the basis of maternal age to match so far as possible the mothers of the mongols. It contains, therefore, an undue proportion of older mothers, but this difficulty can be overcome by standardizing for age. Comparison between these mothers and the mothers of children with congenital dislocation of the hip shows a marked difference in fertility (Table V, overleaf). After standardizing to make the two groups comparable in respect of year of birth of propositi, the mean number of subsequent viable pregnancies was 0·34 per mother for the affected series and 0·43 for the controls. The ratio of these two values (0·79) approximates closely to the figures, obtained earlier, of relative fertility before the birth of propositi.

It seems reasonable to conclude that mothers of children with congenital dislocation of the hip show some impairment of fertility both before and after the birth of the affected child.

Disorders of Pregnancy

Retrospective inquiries into the health of women during pregnancies which resulted in malformed infants are of limited value since it is likely that mothers more readily recall illnesses in pregnancy when the child is abnormal. We have attempted to

TABLE V

FREQUENCY OF PREGNANCIES WHICH OCCURRED AFTER BIRTHS OF PROPOSITI

Maternal Age (yrs) at Birth of Propositus..	Under 25		25–		30–		35–		40 and Over		All Ages	
	Affected	Controls	Affected	Controls	Affected	Controls	Affected	Controls	Affected	Controls	Affected	Controls
No. of Mothers ..	47	14	50	21	38	29	24	39	8	36	167	139
No. of Subsequent Viable Pregnancies by End of 1953	32	10	31	9	4	16	5	14	0	2	72	51
Mean No. of Subsequent Viable Pregnancies per Mother*.. ..	0·68	0·73	0·60	0·31	0·11	0·56	0·22	0·40	0·00	0·05	0·34†	0·43†

* Standardized to the distribution by year of birth of the combined series.
† Standardized to the distribution by year of birth and by maternal age of the combined series.

overcome this difficulty by comparing the present series, not with a sample of births from the general population, but with a Birmingham series of cases of pyloric stenosis which was the subject of an earlier retrospective inquiry (MacMahon, Record, and McKeown, 1951). Pyloric stenosis seemed to be a suitable control since there is evidence that it is dependent much more on post-natal circumstances than on the pre-natal environment (McKeown, MacMahon, and Record, 1951b, 1952). The two series show some differences in the frequency of antenatal disorders (Table VI). This may be due to the small size of the hip series and to the fact that the two groups of mothers were interrogated by different observers. Nevertheless, there is suggestive evidence of an increased liability to toxaemia among mothers of children with hip dislocations. There seems to be no undue prevalence of bacterial or viral disease among the mothers; no instance of rubella was recorded.

TABLE VI

FREQUENCY OF ANTENATAL DISORDERS AMONG PATIENTS WITH CONGENITAL DISLOCATION OF THE HIP OR PYLORIC STENOSIS

Antenatal Condition	167 Patients with Congenital Dislocation of the Hip		488 Patients with Pyloric Stenosis	
	No.	Per cent.	No.	Per cent.
Excessive Vomiting ..	15	9·0	31	6·4
Threatened Abortion ..	5	3·0	19	3·9
Toxaemia	12	7·2	23	4·7
Falls and Other Accidents	8	4·8	37	7·6
Bacterial and Viral Disease	6	3·6	30	6·1

COMPLICATIONS OF DELIVERY

The history of the confinement obtained by retrospective questioning of the mother might also be expected to show some bias, but we believe that, if consideration is restricted to well-defined conditions of which the mother would certainly be told, reliable records can be obtained. It is likely, therefore, that the incidence of the complications listed in Table VII, with the possible exception of "long or difficult labour", are reasonably accurate. Assessment of these results has been facilitated by the publication of data relating to all Birmingham births since 1949 in the Annual Reports of the Medical Office of Health. We used for comparison a single year (1950), but we satisfied ourselves by reference to later reports that this was typical and that the rates did not show a downward secular trend.

TABLE VII

INCIDENCE OF COMPLICATIONS OF BIRTH AMONG 167 MOTHERS OF PROPOSITI AND AMONG ALL BIRMINGHAM BIRTHS IN 1950

Complications of Birth	Mothers of Propositi		All Birmingham Live Births in 1950* (Per cent.)
	No.	Per cent.	
Breech Delivery	27	16·2	3·0
Long or Difficult Labour (Vertex Presentation) ..	20	12·0	—
Twin Delivery	4	2·4	2·7
Placenta Praevia	2	1·2	0·3†
Method of Delivery:			
Forceps (Vertex Presentation)	10	6·0	3·6
Caesarean Section	6	3·6	2·1

* Derived from data published in the Report of the Medical Officer of Health of Birmingham, 1950.
† Based on Birmingham live births in the years 1946–51.

The results confirm the previously reported association of congenital dislocation of the hip with breech delivery. The association is particularly marked in the case of males: excluding cases of Caesarean section, there were 23 males of whom ten (43 per cent.) were delivered by the breech. Of the 138 female births, seventeen (12 per cent.) were breech deliveries. The difference between these proportions is much too large to be reasonably attributed to chance. The difference between the sexes is particularly marked for propositi of Birth ranks

2 and over (Table VIII). Of the thirteen male births in this group, seven (54 per cent.) were breech deliveries; the corresponding figure for females was three in 73 (4 per cent.).

TABLE VIII

FREQUENCY OF BREECH DELIVERY ACCORDING TO SEX AND BIRTH RANK

Birth Rank	Sex	No. of Breech Deliveries	No. of Cephalic Deliveries	Frequency of Breech Delivery (Per cent.)	$\chi^2_{(1)}$	p
1	Male	3	7	30	—0·03	0·9
	Female	14	51	22		
2 and Over	Male	7	6	54	21·95	<0·0001
	Female	3	70	4		
Total	Male	10	13	43	11·56	<0·001
	Female	17	121	12		

Although no comparable figure is available for the general population, the incidence of long or difficult labour (12 per cent.) seems to be higher than one would expect. This impression is supported by the raised frequency of forceps delivery, but assessment of these results is difficult because of the higher proportion of first births among propositi than in the general population.

The number of twin deliveries does not differ appreciably from the expected value. The same conclusion was reported by Idelberger (1951). The incidence of placenta praevia appears to be greater than usual, but numbers are too small to establish a definite association.

There was no obvious difference between bilateral and unilateral cases in the frequency of complications of delivery.

SEASON OF BIRTH

Dates of birth were obtained from hospital records and were therefore available for the whole series of 186 cases. When the seasonal distribution of these cases is compared with that of all maternities in England and Wales for the same years, the number of propositi born in the first and last quarters of the year is greater than expected, and there is a corresponding deficiency of cases in the second and third quarters. The differences are particularly marked in the first and third quarters (Table IX).

In attempting to account for these discrepancies, we may first consider the possibility that the seasonal distribution of births in Birmingham differs from that of the rest of England and Wales. There is evidence from a previous inquiry (McKeown and Record, 1951) that this is not so; for the years 1940–47 the percentage distributions of Birmingham births in the first, second, third, and fourth quarters were respectively 24·9, 25·9, 24·7, and 24·5. These figures do not differ appreciably from the "expected" percentages shown in Table IX.

TABLE IX

NUMBER OF CASES OF CONGENITAL DISLOCATION OF THE HIP ACCORDING TO MONTH OF BIRTH

Month of Birth	No. of Propositi	Percentage Distribution		Difference $O - E$ (per cent.)
		Observed (O)	Expected* (E)	
January February March ..	20 21 22	33·9	25·2	+8·7±3·2
April .. May .. June ..	8 16 15	21·0	26·0	−5·0±3·2
July .. August .. September	11 10 12	17·7	25·0	−7·3±3·2
October.. November December	14 13 24	27·4	23·8	+3·6±3·1
Total ..	186	100	100	

* Based on all maternities in England and Wales, 1942–52.

The second possibility is that, although all births do not show any marked seasonal fluctuation in incidence, first births and later births separately may do so. A series which contained an unusual proportion of first born would then show a seasonal trend. National statistics do not permit a direct examination of this point because there are no published data on the monthly or quarterly incidence of births in relation to parity. There are, however, statistics relating season of birth to maternal age (Annual Reports of the Registrar-General, Pt. II), and these provide useful information, since it may be reasonably assumed that the great majority of mothers under 20 are primiparae and that most women aged 40 and over at the time of the birth are multiparae. These two groups show surprisingly little difference in the monthly distribution of births; in 1950 the proportion of births which occurred in the 6 months from April to September was 50·4 per cent. for women under 20 and 50·3 per cent. for women of 40 and over. It appears, then, that the association of incidence of congenital dislocation of the hip with season of birth is not due to its association with primogeniture. The same conclusion is reached if first and later born propositi are examined separately for evidence of seasonal variation. After excluding nineteen cases of unknown birth rank, there were eighty first born, 32 of whom were born in the period April–September. Of the 87 later born, 34 were born in the same period.

There seems to be little doubt, therefore, that there is some relationship between the incidence of congenital dislocation of the hip and season of birth. This might be due to one or more of a large number of factors, but we here consider only the simplest and most obvious one—temperature. Mean air temperatures, based on Birmingham records for the last 60 years, were obtained from the Annual Reports of the Medical Officer of Health. The relative incidence of congenital dislocation of the hip was calculated for each month by dividing the number of cases born in the month by the number which would have been expected if the condition were independent of season. The trend of these values, shown in Table X, is not perfectly smooth because of the small size of the series, but if April (where the incidence appears to be understated) is excluded, there is a steady gradation from the high values of the winter months to the low figures of July and August. There is obviously a marked negative association between relative incidence and air temperature, but it cannot be decided by inspection of the data whether incidence is correlated most strongly with the temperature of the same month or with that of preceding or succeeding months. This was examined by calculating co-efficients of correlation between relative incidence and the mean air temperature of the same month and of each of the two earlier and of the two later months. For example, the incidence in January was related in turn to temperatures in November, December, January, February, and March. (It may be objected that the relationship between incidence and temperature is not linear, and in fact, the regression is much better expressed by a quadratic curve. The use of the correlation coefficient in these circumstances is justified by the fact that it is used here only as a comparative index.) The results (Table XI) suggest that incidence in any month is related more closely to the temperature of the same month than to that of the preceding or subsequent months. It is possible, however, that the abnormally low incidence in April may have a disproportionate influence on the correlations and it seemed advisable to repeat the calculations after excluding cases born in this month. This procedure gives slightly closer correlations, and the temperature in the month before birth seems to be at least as important as that of the month of birth.

TABLE X

RELATIVE INCIDENCE OF CONGENITAL DISLOCATION OF THE HIP AND MEAN AIR TEMPERATURE ACCORDING TO MONTH OF BIRTH

Month of Birth	Relative Incidence	Mean Air Temperature (°F.)
January	1·28	38·8
February	1·43	39·2
March	1·33	42·0
April	0·50	46·3
May	0·96	52·2
June	0·95	57·4
July	0·69	60·8
August	6·66	60·1
September	0·79	56·0
October	0·94	49·7
November	0·91	43·0
December	1·59	39·9

TABLE XI

CORRELATION BETWEEN MONTHLY RELATIVE INCIDENCE AND MEAN AIR TEMPERATURE OF EARLIER, SAME, AND LATER MONTHS

Month of Temperature Record relative to Month of Birth	Coefficient of Correlation	
	All Cases	Excluding Cases Born in April
2 Months Earlier	− 0·35	− 0·63
1 Month Earlier	− 0·64	− 0·89
Same Month	− 0·72	− 0·87
1 Month Later	− 0·65	− 0·67
2 Months Later	− 0·41	− 0·31

The influence of temperature may also be examined by relating secular change in incidence to the prevailing temperatures. Considering first the 3 months January to March, the mean quarterly air temperature was unusually low in the years 1942, 1947, and 1951, during which 22 propositi were born; the three years with the highest temperatures in this quarter were 1943, 1948, and 1950, which yielded sixteen cases. The coefficient of correlation between the number of cases in this quarter in each of the eleven years 1942–52 and mean air temperatures was −0·25. The second quarter (April–June) showed a similar difference in the number of cases between the 3 years with the lowest temperatures and the three with the highest (sixteen and ten respectively); the coefficient of correlation based on the 11 years was −0·64. The third quarter showed a slight difference in the opposite direction (seven and nine cases) and a slight positive correlation (+0·17), but in the fourth quarter the influence of temperature was again evident: fifteen propositi were born in the three coldest years and nine in the warmest, and the correlation coefficient was −0·31.

In view of the small number of observations, little importance can be attached to the magnitude of these coefficients, but it seems reasonable to suppose that the sign has some significance. The negative correlation between incidence and air temperature during the winter, spring, and autumn seasons suggests that low temperature may be of aetiological

importance. The trivial positive correlation during the warmest quarter of the year (July to September) is not necessarily inconsistent with this view, since the temperature at that time may rarely fall low enough to have any influence.

There was some indication that males were more sensitive to seasonal influences than females. Of the 27 males in the series, twenty (74 per cent.) were born in the period from October to March; the corresponding figure for females was 94 out of 159 (59 per cent.). The relative frequency of bilateral and unilateral dislocations did not appear to vary according to season of birth.

Familial Incidence

(*a*) *Twins.*—There were two male and two female propositi who were twins, all from like-sexed pairs. Dislocation of the hip was diagnosed in only one of each pair, but one unaffected twin (female) showed radiographic evidence of acetabular dysplasia. An unaffected member of one of the male pairs was reported to have talipes. The large series of twins reported by Idelberger (1951) did not show an unusual incidence of defects in the unaffected members.

(*b*) *Younger Sibs.*—In the case of families containing two or more affected children, later sibs were enumerated from the first propositus and not from the first affected child if this was not a propositus. After excluding children who had died or were less than 3 years old at the time of the survey, there were 25 younger brothers, none of whom was affected, and 37 younger sisters of whom three were affected (Table XII). These figures are too small to permit more than a rough evaluation of the chance of a subsequent child being affected, but they suggest that the average risk may be about 5 per cent. The actual risk depends, of course, on the sex of the child; possible values are 1 to 2 per cent. for boys and 5 to 10 per cent. for girls. These estimates may vary with season of birth. Vogel's and Isigkeit's data suggest similar risks to siblings of affected. An estimate of 17 per cent. (Reed, 1955) appears to be too high.

(*c*) *Older Sibs.*—There were 81 older brothers, one of whom had congenital dislocation of the hip, and 79 older sisters, seven of whom were affected. The incidence in prior sibs was, therefore, very similar to that in subsequent sibs.

(*d*) *Parents.*—There were five affected mothers. No fathers were affected. No history of consanguineous marriage was obtained. These data do not, of course, permit an assessment of the risk to children of an affected parent, but if it is assumed that the incidence has not changed and that the fertility of affected persons is not grossly impaired, it seems likely that this risk is not greater than that in children born after an affected sibling.

(*e*) *Other Relatives.*—Inquiry was made into the occurrence of congenital dislocation of the hip among siblings of the parents (uncles and aunts of propositi) and among their children (first cousins of propositi). A few mothers had only a vague knowledge of their nephews and nieces and even less of their husband's relatives. In these circumstances estimates of incidence might be liable to bias and a comparison of incidence between paternal and maternal relatives might give misleading results. We have attempted to overcome this difficulty by considering only those relatives whom the mother could name, since it seems likely that if she knew the name she would know whether or not the individual had a dislocation. Children who had died in infancy and those too young to manifest the condition were excluded. Care was taken to avoid duplication of an affected relationship; for example, if there were two propositi who were cousins only one was counted as an affected relative. Results are summarized in Table XII (overleaf).

Because of the low frequency of the condition in relatives, reliable figures of incidence cannot be obtained from a series of this size, but some broad conclusions are possible. Considering all relatives together, incidence does not appear to vary with the sex of the propositus. As expected, female relatives are affected much more frequently than males, and the proportion of males (9 per cent.) is not very different from the usual sex ratio. The incidence is considerably greater among sibs than among parents, and among cousins than among uncles and aunts. There is no indication that the disability is transmitted more frequently through the distaff side of the family.

The incidence of other malformations among relatives is of considerable interest. It was considered that adequate details of these defects among uncles, aunts, and cousins of propositi could not be expected, and the examination has been restricted to incidence among siblings. Excluding children who had congenital dislocation of the hip, there were 250 live-born siblings, of whom nine (3·6 per cent.) had congenital defects (Table XIII, overleaf). This incidence is greater than that for a representative series of live births (2·9 per cent.) reported by Grundy and Lewis-Faning (1957). The difference appears to be due to the prevalence of defects which are sometimes associated with congenital dislocation of the hip (talipes, torticollis, and hernia).

TABLE XII

INCIDENCE OF CONGENITAL DISLOCATION OF THE HIP AMONG RELATIVES OF PROPOSITI

Propositi	Relatives: Relationship to Propositus	Male: Total No.	Male: No. Affected	Female: Total No.	Female: No. Affected	Both Sexes: Incidence per 1,000
Male ..	Younger Sibs	5	0	4	0	7
	Older Sibs	9	0	10	1	
	Parents	25	0	25	2	
	Uncles or Aunts (Paternal)	46	0	40	0	
	Uncles or Aunts (Maternal)	48	0	51	0	
	Cousins (Paternal)	22	0	33	0	
	Cousins (Maternal)	66	0	55	0	
Female ..	Younger Sibs	20	0	33	3	9
	Older Sibs	72	1	69	6	
	Parents	139	0	139	3	
	Uncles or Aunts (Paternal)	223	0	201	1	
	Uncles or Aunts (Maternal)	232	1	251	0	
	Cousins (Paternal)	174	0	186	2	
	Cousins (Maternal)	239	0	250	2	
		Incidence per 1,000		Incidence per 1,000		
Both Sexes	Younger Sibs	0		81		48
	Older Sibs	12		89		50
	Parents	0		30		15
	Uncles or Aunts (Paternal)	0		4		2
	Uncles or Aunts (Maternal)	4		0		2
	Cousins (Paternal)	0		9		5
	Cousins (Maternal)	0		7		3
	Total	2		15		8

TABLE XIII

FREQUENCY OF MALFORMATIONS AMONG 250 LIVE-BORN SIBLINGS (EXCLUDING CASES OF CONGENITAL DISLOCATION OF THE HIP)

Malformation	No.	Per cent.
Talipes	2	1·6
Torticollis	1	
Diaphragmatic Hernia	1	
Spina Bifida	1	2·0
Hare-lip and Cleft Palate	1	
Pyloric Stenosis	1	
Hydrocele	1	
Polydactyly	1	
Total	9	3·6

DISCUSSION

The term "congenital dislocation of the hip" has continued since it was originated by Hippocrates, although its literal meaning is misleading. "Congenital" (ἐκ γενεης) is inappropriate, in that the dislocation itself is probably present at birth in only a minority of cases, and "dislocation" (ἐξαρθρησις) conveys an implication of abruptness. In the dislocation of the infantile hip the gradual onset and the intact capsule suggest that the process is usually the result of a gradual displacement brought about by differential growth under abnormal pressures and has more in common with deformities induced by bad posture than those which result from sudden forces.

Before attempting to formulate a hypothesis it may be useful to recapitulate the more important facts based on evidence derived from this and earlier inquiries:

(1) There is a marked geographical variation in incidence.

(2) The condition is much more frequent among females than males.

(3) The left hip is involved more frequently than the right.

(4) Associated defects are common. They usually involve the musculo-skeletal or nervous systems. These musculo-skeletal defects are also more frequent among siblings.

(5) The defect occurs more commonly among first-born than among later-born, and is often associated with breech delivery.

(6) Incidence is greater in children born in winter than in those born in summer and seems to be inversely related to air temperature.

(7) There is a marked tendency for the condition to run in families. Maternal and paternal relatives appear equally liable to the defect.

(8) Twin studies indicate that the foetal genotype considerably influences but does not necessarily determine the development of dislocation.

The evidence strongly suggests that both genetical and environmental factors are important. The circumstances after conception which comprise the environment may conveniently be divided into those operating before birth, during birth, and after birth, but it is not always possible to decide with certainty when a particular factor operated. As knowledge of the environment before birth is largely based on observations made at birth, and as the contribution of the foetal genotype to such circumstances as foetal lie and quantity of liquor amnii is unknown, it is difficult to obtain any information implicating the pre-natal environment which cannot be interpreted as evidence relating either to genetic factors or to parturition. But there is no firm evidence that the actual conditions of birth have any influence, and this view is supported by the observation that incidence is not reduced in children delivered by Caesarean section.

The greatly increased risk associated with primogeniture may reasonably be accepted as evidence of some features of the uterine environment present at first, but not at later, births which are sufficient to double the average liability to dislocation.

The considerable variation in liability to dislocation with season of birth suggests that the differences between summer and winter in England may be sufficient to lead to a doubling of the risk, and even greater seasonal variations have been observed in Japan (Mizuno, 1957). In attempting to explain this seasonal effect, we may first consider the possibility that it is due to variations in maternal diet. There is no doubt that there are seasonal changes in the character of the diet and particularly in its vitamin content, but it seems unlikely that this can account for the seasonal incidence of congenital dislocation of the hip as there is no evidence that the condition is commoner among malnourished populations or is associated with deficiency diseases. Another possibility is that the association may be due to the effect of temperature on the tone of the uterus and abdominal muscles. We know of no direct evidence that temperature influences the tone of the uterine musculature and it is likely that its effect on the tone of the abdominal muscles is trivial compared with the effect of previous pregnancies. The fact that the influence of season on incidence is as marked as that of parity suggests, therefore, that this explanation is not adequate. Lastly, we may consider factors in the post-natal environment which may account for this variation of incidence with season. The influence of cold in increasing the muscle tone of the infant can probably be dismissed because it would have, if anything, a protective action. The most likely explanation is that infants born in winter, particularly during cold weather, have to endure heavier clothing and cot coverings. This is likely to cause severe restriction of limb movement and prevent the thighs from assuming the natural position of flexion, abduction, and external rotation which is similar to the position adopted in the conventional treatment of the deformity. Support for this view is provided by the extremely high incidence reported among some Red Indian and Lapp communities; this may be related, in part, to the constrictions imposed by the shoe-like Red Indian cradle into which the infant is laced, and the coffin-like Lappic cot.

Evidence relating to the influence exerted by the environment after birth has not previously been seriously considered and may be received with scepticism. It is, however, no more surprising that variation in the infant's environment should influence any predisposition to dislocation than that the environment imposed by the surgeon's plaster spica should be effective in reversing this process.

We may now consider the mechanism by which genetic and environmental agencies may predispose to dislocation. This may be brought about by:

(1) Variations in obliquity, eccentricity, or depth of the acetabulum.

(2) Variations in the forces of approximation between the head of the femur and the acetabulum; these will be determined mainly by the muscle tone of the foetus and infant.

(3) Variations in the other forces acting on the joint surfaces. These will be determined mainly by the position of the legs, and by the compressive forces acting on the legs of the foetus or infant, either from the uterine wall before birth or from clothing after birth.

These three groups may, of course, interact with one another. A possible form of interaction, advanced by Browne (1957), is that the application of pressure between the head of the femur and the acetabular rim may sometimes be so great as to interfere with normal growth, leading to a self-perpetuating condition of acetabular dysplasia. This concept of pressure dysplasia had previously been advanced in relation to infantile scoliosis and club foot (Browne, 1956) and probably has wide application. It is more readily appreciated if it is realized that the distance through which dislocation occurs is small compared to the change in size of the parts with growth. Although a dislocation cannot develop *de novo*, since the whole hip joint evolves from a single mass of cartilage, it could easily develop as a consequence of differential growth of the various parts without stretching or rupture of any part.

Evidence bearing on these points is provided by consideration of abnormalities associated with congenital dislocation of the hip. They may be classified into six groups:

(1) Conditions leading to gross neuro-muscular disturbances. Spina bifida, amyotonia, arthrogryposis, and cerebral diplegia are not infrequently associated with congenital dislocation of the hip. Spina bifida is a particularly common precursor, although the association will usually escape notice unless the hip joints are opened at autopsy.

(2) Conditions predisposed to by a shallow, oblique, or eccentric acetabulum. Perthes' disease and osteo-arthritis of the hip are often associated with these variations from the normal, and the incidence of both is raised among relatives of persons with congenital dislocation of the hip (Isigkeit, 1928).

(3) Conditions leading to generalized or localized impairment in muscle tone or co-ordination. These conditions might be expected to result in club-foot, torticollis, scoliosis, and herniae, all of which are found more frequently than usual, not only in persons with congenital dislocation of the hip (Poli, 1937) but also in their siblings and other relatives (Storck, 1938; Isigkeit, 1928). Such muscular weakness or incoordination might also predispose to inability of the foetus to assume a vertex presentation, either through inactivity or through failure to fold the legs (Vartan, 1940).

(4) Conditions predisposed to by increased uterine pressure. Increased uterine pressure could explain most, if not all, of the associated abnormalities considered in the previous group, and also the tendency to breech delivery (Vartan, 1950). The high incidence of such abnormalities in paternal relatives and the results of twin studies limit the adequacy of this explanation. Browne (1957) considers that there is a distinct sub-group of cases of hip dislocation associated with club foot which is readily explained by extreme uterine pressure. Our data on nine cases with these combined abnormalities do not show any remarkable distribution by birth rank or method of delivery. Increase in uterine tone might also account for the impaired rate of foetal growth and perhaps for the increased incidence of toxaemia noted in this series.

(5) Conditions predisposed to by persistent breech presentation. Torticollis appears to be the only other common malformation to which breech delivery particularly predisposes (Storck, 1950). We were not able to confirm Storck's view that a pronounced tendency to persistent breech presentation is genetically determined and is mainly responsible for the hereditary tendency to congenital dislocation of the hip. Our data on breech delivery in siblings did not suggest that it was more frequent than in the general population. The Julio-Claudian family provides an interesting example of a familial association of obstetric difficulties, including breech delivery, and lameness.

(6) Other congenital abnormalities. These do not appear to occur with increased frequency among children with congenital dislocation of the hip and the association may be regarded as fortuitous. Poli's observations are consistent with this interpretation of our more limited data.

Any attempt to postulate an acceptable genetic hypothesis for congenital dislocation of the hip must be made in cognizance of the fact that environmental variations may greatly increase the liability of manifestation, and that the hip is a structure of such variability and complexity that it would seem wholly unrealistic to expect that such a deformity should be determined by a single gene substitution. The fact that the pelvis shows appreciable sexual differences, even as early as the third month of foetal life (Dega, 1933) suggests that it is not necessary to postulate sex-linked genes.

Because of the large number of genetical and environmental factors which appear to influence the liability of the hip to dislocate, the degree of stability of the articulation in a population of new-born infants would be expected to show continuous variation. The imagined distribution of a series of hip joints is shown in Fig. 2 (opposite), the more stable joints being on the left and the more labile on the right.

Let us suppose that such a series is exposed to varying degrees of deforming force, represented by the dotted line in the figure. Joints indicated by the area ABB′ would never dislocate, however unfavourable the postnatal environment. Those distributed near to the line CC′ would be dislocated only by very unfavourable circumstances, and those near to the line DD′ would usually dislocate. The most labile joints, represented by the area EFE′, would always dislocate, however gently the infant were handled, and would frequently be dislocated before birth.

The orientation of the frequency distribution relative to the horizontal axis depends on a number of factors. There is evidence that, because of differences in the depth and inclination of the acetabulum, the hip joints of Sudanese and possibly of most indigenous African peoples are stronger than those of white races (Cheynel and Huet, 1952).

Fig. 3 (opposite) shows hypothetical distributions of the hip joints of two populations, the solid line

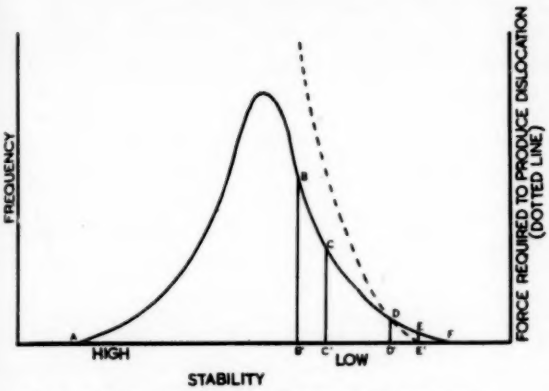

FIG. 2.—Hypothetical distribution of hip-joints according to stability.

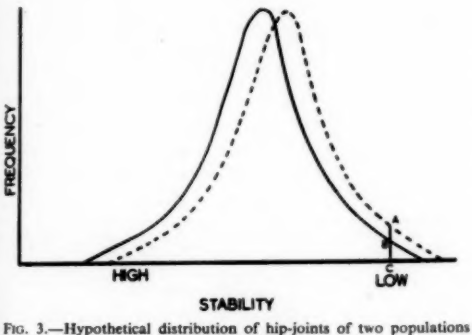

FIG. 3.—Hypothetical distribution of hip-joints of two populations according to stability.

representing an African and the dotted line a white population. The application of a given stimulus (ABC) to the joints of both groups would clearly cause more dislocations among the whites than among the Africans.

The same sort of model may be used to account for the differences in incidence between males and females (males being represented by the solid line and females by the dotted line), and between the two sides (solid line for the right side and dotted line for the left). These differences are supported by observations based on measurements of the hip joints of one hundred foetuses reported by Dega (1933).

It appears, then, that the hypothesis examined here is not inconsistent with the more important points known about congenital dislocation of the hip. It is possible that the general terms of the hypothesis are more widely applicable and may help in understanding other defects. We have, at present, no conclusive evidence that this is true of other human malformations, but there are indications that some animal abnormalities can be explained in this way. Grüneberg (1951) has described a foramen in the acetabulum of the mouse apparently determined by multifactorial inheritance and showing what he terms quasi-continuous variation. Other abnormalities in mice inherited in a similar way have been shown to be influenced by the uterine environment (Deol and Truslove, 1957). The crooked-toe defect of poultry is considered by Hicks (1953) and Lerner (1954) to be the expression of one tail of a continuous distribution determined by multifactorial inheritance. Manifestations of the condition is influenced by sex and by such environmental factors as walking on wire-netting. Lerner also considers that certain malformations of the wings of the fruit fly investigated by Goldschmidt (1945) and Goldschmidt, Hannah, and Piternick (1951) can be explained in a similar way. All these malformations are associated with impairment in fertility.

The hypothesis may be criticized, as other polygenic theories have been, on the grounds that it cannot fail to agree with all observations relating to the inheritance of the disease. Nevertheless, it seems to be more realistic than hypotheses which involve only one or two genes. It may also be of more practical value because it does not divert attention from environmental influences. In the present state of knowledge it is questionable whether eugenic counselling is justifiable, and it would probably be more useful to attempt to control environmental influences by the early recognition and rectification of foetal malpositions and by permitting the infant full limb movement and the adoption of a natural position. It seems quite certain that the advising of parents with an affected child not to reproduce, even if the advice were acted upon, would have a negligible influence on the incidence of the disease. Affected parents do not appear to be responsible for more than about 5 per cent. of cases. A form of advice which would be at least harmless, and might reduce the risks by as much as a half, would be to suggest the late autumn as the most propitious time for conception.

SUMMARY

(1) An attempt was made to ascertain all children who were born in the period 1942–52 and were treated in Birmingham hospitals for congenital dislocation of the hip. The incidence was 0·65 per

1,000 live births. Further information on 167 patients was obtained by interviewing the mothers.

(2) The data were consistent with previous reports relating to the sex ratio (15 per cent. males) and the side more commonly affected (64 per cent. of affected hips were left-sided). The frequencies of associated defects (16 per cent.), and of breech delivery (17 per cent.) were rather higher than in most previous reports.

(3) The mean birth weight was about $\frac{1}{2}$ lb. below the expected value. This could not be explained by any obvious decrease in the duration of gestation.

(4) The incidence among first-born was about twice that in second and third children. There appeared to be some association with increasing maternal age, and there was some evidence that the fertility of mothers was slightly impaired.

(5) The incidence among children born in the winter was double that of children born in the summer. Examination of annual variations suggested that incidence was related to air temperature.

(6) The risk of the condition occurring in a subsequent child appears to be about 5 per cent., but is dependent on the sex of the child and possibly on the season of birth.

(7) Current hypotheses are considered in relation to these findings. It is suggested that the stability of the hip joint is determined by multifactorial inheritance and by a number of factors in the pre-natal and post-natal environment. The most important of these are considered to be those which influence the attitude and activity of the foetus and of the infant.

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INVESTIGATION OF THE MEDICAL AND SOCIAL NEEDS OF PATIENTS IN MENTAL HOSPITALS

II. TYPE OF ACCOMMODATION AND STAFF REQUIRED

BY

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A previous communication (Garratt, Lowe, and McKeown, 1957) described an investigation of the medical and social needs of the 3,555 Birmingham residents in Birmingham mental hospitals in January, 1957. Reasons were given for believing that these patients were representative of the mental hospital population of England and Wales, and the patients were classified according to the type of facilities required for their care. It was considered that 13 per cent. needed the full resources of a hospital, 75 per cent. needed limited hospital facilities, and the remaining 12 per cent. required none of the services traditionally associated with hospitals. The purpose of the present report is to examine in greater detail the patients assigned to each class, and to suggest the type of accommodation and staff required.

The methods used to obtain the data were described fully in the first communication. Briefly, with the help of the psychiatrist responsible for his care, the needs of each patient were examined and used to assess the nature of the services which would be most suitable.

I. Patients Needing Full Hospital Facilities

It was estimated that 459 (13 per cent.) of the 3,555 Birmingham patients in mental hospitals needed to be cared for in an institution which could provide the full range of services associated with a modern hospital: *x*-rays, laboratories, surgery, skilled nursing, etc. There is of course, some room for differences of opinion about the type of service which necessitates full hospital care. In the case of patients at home, for example, decision turns upon the availability of out-patient departments, day-hospitals, laboratories, etc. The problem is different when patients are already in hospital, since it is then somewhat unrealistic—particularly in the absence of knowledge of their home circumstances—to inquire whether they need have been admitted. All that was attempted was to assess whether their care required access to full hospital facilities. Undoubtedly there were a few patients who, if their homes had been suitable, could have been treated as out-patients.

(*a*) Type of Patient

Table I gives the distribution of the 459 patients according to the nature of their mental illness, based on the 1948 International Statistical Classification. The distribution is very similar to that of all 3,555 patients in the mental hospitals (Garratt and others, 1957: Table V); the most notable differences are the lower incidence of senile psychosis (4·4 per cent. compared with 13·4 per cent.) and the higher incidence of psychoneurosis (7·4 per cent. compared

TABLE I

DIAGNOSIS OF PATIENTS WHO REQUIRED FULL HOSPITAL FACILITIES

Diagnosis*	Males	Females	All Patients	
			Number	Per cent.
Schizophrenia (300) ..	128	71	199	43·4
Manic Depressive Reaction (301)	30	32	62	13·5
Involutional Melancholia (302)	15	29	44	9·6
Paranoid State (303) ..	18	15	33	7·2
Senile Psychosis (304) ..	6	14	20	4·4
Other Psychoses (305–09)	11	18	29	6·3
Psychoneuroses (310–18) ..	10	24	34	7·4
Disorders of Character and Behaviour (320–24; 326)	12	—	12	2·6
Mental Defect (325) ..	2	5	7	1·5
Syphilis (025)	—	2	2	0·4
Epilepsy (353)	6	8	14	3·1
Other Disorders	1	2	3	0·7
Total	239	220	459	100

* Numbers in brackets are list numbers used in 1948 International Statistical Classification.

with 1·9 per cent.). There is also a considerable difference between the sex ratios, the percentage of males being 52·1 and 38·2 among the 459 and 3,555 patients respectively. The excess of males among patients considered to require full hospital care is due chiefly to the sex ratio of schizophrenics (128 males, 71 females) and of patients with disorders of character and behaviour (12 males, 0 females). A preponderance of males has also been noted among recent hospital admissions for England and Wales as a whole (Registrar-General, 1955), but the reason for it is obscure. Broadly, however, the patients who were thought to require full hospital care were not restricted to a small group of diseases, but were widely selected from those represented in the mental hospitals.

It is well known that there are in mental hospitals some patients who, in addition to their mental illness, have a physical illness which, at least for a time, determines the character of the care they require. We have therefore divided the 459 patients needing full hospital care (Table II) according to whether their need was determined by their mental condition (82·8 per cent.) or by a superimposed physical illness (17·2 per cent.). All of the physically ill patients either were also mentally ill, or had had a mental illness at the time of their admission to hospital—otherwise of course they would not have been in a mental hospital. But at the time of the inquiry their needs were largely determined by their physical condition, and were therefore sharply identified from those of the main body of patients considered to require full hospital care.

TABLE II

TYPE OF ILLNESS FOR WHICH PATIENTS REQUIRED FULL HOSPITAL FACILITIES

Full Hospital Facilities Required for:	Males		Females		All Patients	
	No.	Per cent.	No.	Per cent.	No.	Per cent.
Mental Illness ..	205	85·8	175	79·5	380	82·8
Physical Illness ..	34	14·2	45	20·5	79	17·2
Total	239	100	220	100	459	100

The types of mental illness for which the 380 patients were thought to require full hospital care were approximately as suggested by Table I; indeed, because these patients comprise more than four-fifths of the total in the Table, they largely determine the nature of the distribution. It follows that, as noted in respect of all patients in Table I, they are widely selected from the 3,555 patients in Birmingham mental hospitals, differing chiefly in the lower incidence of senile psychosis, and the higher incidence of psychoneurosis. They are also identified from the total population of the mental hospitals by a higher proportion of voluntary admissions (46·8 per cent. compared with 31·2 per cent.) and of recent admissions (the percentages of patients in hospital for less than a year were 68·2 and 20·0 respectively). Of the 79 patients undergoing investigation and treatment for physical illness, 26 had infectious diseases, 31 other medical conditions, and 22 a variety of disorders needing surgical attention (Table III).

TABLE III

TYPE OF PHYSICAL ILLNESS FOR WHICH PATIENTS REQUIRED FULL HOSPITAL FACILITIES

Type of Illness		Number of Patients
Infectious Conditions	Pulmonary Tuberculosis ..	23
	Other	3
Other Medical Conditions	Cardio-vascular Disease ..	7
	Dermatological Conditions ..	8
	Acute Bronchitis	4
	Other	12
Surgical Conditions	Traumatic and Orthopaedic ..	11
	Malignant Disease	8
	Other	3
Total		79

(*b*) PATIENTS' NEEDS

Medical Treatment.—Table IV (opposite) shows the medical treatment required by those needing full hospital care because of mental illness. (It should perhaps be noted once again that in this discussion the medical treatment required was the treatment actually received, because psychiatrists were able to provide what they considered necessary). There were considerable differences of opinion about the treatment of choice. For example, in one hospital 14·6 per cent. were having E.C.T. and 60·4 per cent. intensive administration of tranquillizers. In another hospital, 63·8 per cent. were having E.C.T. and none was receiving intensive tranquillizers. Our concern here is not, however, with the type of treatment, but with the proportion of patients requiring treatment which would make it desirable to have access to full hospital services. This proportion was 12, 11, 8, and 11 per cent. in the four hospitals. It is perhaps surprising that there were only trivial differences, since there was considerable variation in the composition of the hospital populations.

The medical treatment of the patients who required hospital care primarily because of physical illness (Table III) was quite different. Their illnesses were those usually treated in general hospitals,

TABLE IV

TREATMENT NEEDED BY PATIENTS WHO REQUIRED FULL HOSPITAL FACILITIES FOR MENTAL ILLNESS

Treatment Needed	Investigation and Assessment		Electro-convulsive Therapy		Insulin Therapy		Intensive Tranquillizers		Other		Total	
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Hospital A	34	23·6	21	14·6	1	0·7	87	60·4	1	0·7	144	100
Hospital B	39	37·5	44	42·3	2	1·9	19	18·3	—	—	104	100
Hospital C	33	44·6	28	37·8	7	9·5	1	1·4	5	6·8	74	100
Hospital D	19	32·8	37	63·8	2	3·4	—	—	—	—	58	100
All Hospitals	125*	32·9	130	34·2	12	3·2	107	28·2	6†	1·6	380	100

* 32 of the 125 patients under investigation and assessment were receiving sedatives, vitamins, amphetamine, etc.
† Four patients were receiving psychotherapy and two penicillin for general paralysis of the insane.

where all the necessary facilities exist, and they were in mental hospitals only because they were then, or at admission, also mentally ill. Nevertheless it was their physical condition which, for a time at least, determined the accommodation and staff which they needed. In most cases the onset of the physical illness had occurred while the patient was in a mental hospital, but some patients had been transferred from a general hospital and a considerable number (23) had been admitted directly from home (Table V).

TABLE V

LOCATION OF PATIENTS, WHO REQUIRED FULL HOSPITAL FACILITIES FOR PHYSICAL ILLNESS, AT THE ONSET OF THAT ILLNESS

Location	Home		Mental Hospital	Total
	Direct Admission	Transfer from General Hospital		
Number	23	3*	53	79
Per cent.	29·1	3·8	67·1	100

* One patient was suicidal, and two had been extremely confused and restless.

Nursing Care.—Table VI gives the results of assessment of nursing care. All of the patients treated for mental illness needed skilled nursing, and 90 per cent. needed it regularly (at least once a day) or constantly. The small proportion of patients who needed only occasional skilled nursing were in the late stages of their treatment, and had had more frequent attention a short time earlier. Since all of these patients were undergoing investigation or treatment for a mental illness, the service they required was that of a skilled mental nurse. Relatively few of them (13·2 per cent.) required simple basic nursing, and in this respect their needs were sharply distinguished from those of the patients treated for physical illness. This difference is of course determined by the fact that almost all of them were fully ambulant (91·3 per cent.: Table VII) and continent (95·3 per cent.: Table VIII). They did not therefore require the frequent personal attention needed by bedfast patients.

The demands for nursing care of patients with physical illnesses (Table VI) were quite different. Again, all of them required skilled nursing and, except for the few in the late stages of treatment, they required it so frequently that it could scarcely have been provided outside hospital. But the type of skilled nursing was different, being essentially of the kind needed by patients in general hospitals. It is in

TABLE VI

NURSING CARE NEEDED BY PATIENTS WHO REQUIRED FULL HOSPITAL FACILITIES

Full Hospital Facilities Required for:	Nursing Care Needed	Constant		Regular*		Occasional		None		All Patients	
		No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Mental Illness	Skilled	13	3·4	340	89·5	27	7·1	—	—	380	100
	Basic	9	2·4	37	9·7	4	1·1	330	86·8	380	100
Physical Illness	Skilled	6	7·6	59	74·7	14	17·7	—	—	79	100
	Basic	26	32·9	36	45·6	5	6·3	12	15·2	79	100

* At least once a day.

TABLE VII

MOBILITY OF PATIENTS WHO REQUIRED FULL HOSPITAL FACILITIES

Full Hospital Facilities Required for:	Bedfast		Sitting Out of Bed		Ambulant				All Patients	
					Partly		Fully			
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Mental Illness ..	10	2·6	12	3·2	11	2·9	347	91·3	380	100
Physical Illness ..	51	64·6	10	12·7	6	7·6	12	15·2	79	100

respect of basic nursing, however, that the demands of the two groups of patients differ most sharply. Because a high proportion of the physically ill patients were confined to bed (Table VII) and because many more of them were incontinent (Table VIII), their need for simple personal nursing was far greater.

TABLE VIII

CONTINENCE OF PATIENTS WHO REQUIRED FULL HOSPITAL FACILITIES

Full Hospital Facilities Required for:	Incontinent				Continent		All Patients	
	Wholly		Partly					
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Mental Illness	11	2·9	7	1·8	362	95·3	380	100
Physical Illness	18	22·8	2	2·5	59	74·7	79	100

More than three-quarters of them required regular or constant service of this kind (Table VI), and relatively few (15·2 per cent.) were independent of it.

Supervision.—Another feature which has a considerable bearing upon the type of accommodation and staff is the degree of supervision required because of mental illness. Patients were subdivided according to whether they needed close supervision (personal supervision of severely disturbed patients), limited supervision (supervision of patients who, although they did not require close supervision, could not be allowed full freedom within the hospital community), open supervision (confinement to hospital but with complete freedom of movement within it), or no supervision. The proportions of patients in each class are given in Table IX. It is scarcely surprising that all the patients undergoing active investigation and treatment for mental illness were thought to require some supervision; nevertheless two-thirds of them were restricted only insofar as they were not allowed alone outside the hospital grounds. In some respects supervision was less significant in the case of patients treated for physical illness because many were confined to bed or were only partly ambulant, and the possibility of unrestricted movement scarcely arose, Nevertheless, 11·3 per cent. of the patients required close supervision and a further 78·5 per cent. required limited supervision on the wards. It seems clear that although the medical and nursing care was determined by their physical condition, because of their associated mental illness most of these patients would not be suitable to be admitted to the same wards as patients in general hospitals with similar physical illnesses, but without the added complication of mental illness. And, although the skilled nursing was mainly directed to the care of physical illness, it could best be provided by nurses with experience of mental as well as of physical illness. It should also be noted that, after completion of treatment of their physical illness, relatively few (7) of these 79 patients were likely to be able to return home. It was thought that most of them (66) would require to live in an institution which could provide simple care of mental illness (of the type referred to under "limited hospital facilities") while the remainder (6) would need a further period of access to full hospital facilities for the treatment of their mental illness.

TABLE IX

SUPERVISION NEEDED BY PATIENTS WHO REQUIRED FULL HOSPITAL FACILITIES

Full Hospital Facilities Required for:	Supervision								All Patients	
	Close		Limited		Open		None			
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Mental Illness ..	9	2·4	117	30·8	254	66·9	—	—	380	100
Physical Illness ..	9	11·3	62	78·5	—	—	8	10·1	79	100

(*c*) ACCOMMODATION REQUIRED

It seems to us that the two considerations which have most bearing on the type of accommodation and staff which should be provided for mentally-ill patients who need the full range of hospital facilities are their mobility, and the degree of supervision required. Unlike patients in general hospitals, most of them are ambulant and do not need traditional ward accommodation. The degree of supervision has a considerable bearing upon the type of nursing. For it might be possible to use nurses trained or in training for the general register for care of patients who require little or no supervision, but it would probably not be consistent with present nursing policy to use them in units containing seriously disturbed patients.

In Table X the 459 hospital patients have been classified according to their mobility and need for supervision. In this Table we have also preserved the distinction between patients treated for mental illness and those treated for physical illness. But, although this distinction has a considerable bearing on care, it is not a precise guide to the most suitable type of accommodation and staff. This is because the physically-ill were by no means all bedfast patients who needed no supervision, and the mentally-ill were not all ambulant patients who needed supervision. But by combining certain of the cells in Table X we have obtained four groups of patients identified broadly according to supervision and mobility.

(A) Bedfast, or recently bedfast, patients, all of whom required some degree of supervision.

(B) Bedfast, or recently bedfast, patients who required no supervision.

(C) Ambulant patients restricted—because of their mental state—within the hospital community.

(D) Ambulant patients unrestricted within the hospital community, but not allowed outside the hospital grounds.

It is suggested that the four groups of patients identified in this way should be accommodated in four different types of unit (Table XI). They should be in either traditional ward or hostel type of accommodation, and, according to the supervision required, in a closed or an open block. It should perhaps be said that the term "open block" has a different significance in the two cases. The eight patients (Group B) suitable for a traditional and open ward required no mental supervision whatsoever; their needs were identical with those of patients in general hospitals, and there could have been no reasonable objection either to admitting them to general wards, or to using student nurses in training for the general register for their care. The 241 patients suggested for hostel-type accommodation in an open block (Group D) all required some supervision because of their mental state, although they could move freely within the hospital community. These differences are of course evident from the more detailed classification given in Table X.

TABLE XI

TYPE OF ACCOMMODATION NEEDED BY PATIENTS WHO REQUIRED FULL HOSPITAL FACILITIES

Accommodation	Traditional Ward	Hostel-Type	Total
Closed Block	(A) 93	(C) 117	210
Open Block	(B) 8	(D) 241	249
Total	101	358	459

Ward Accommodation with Supervision (Group A).—In Group A there are 93 patients (20 per cent.

TABLE X

CLASSIFICATION OF PATIENTS WHO REQUIRED FULL HOSPITAL FACILITIES

Patients		Bedfast or Sitting Out of Bed		Partly or Fully Ambulant		All Patients
Illness		Mental only	Physical	Physical	Mental only	
Supervision ..	Close	(A) 1	8	1	(C) 8	18
	Limited	8	47	15	109	179
	Open	13	—	—	(D) 241	254
	None	—	(B) 6	2	—	8
Total		22	61	18	358	459

of 459), who were considered to need traditional ward accommodation with supervision. 71 of them were being treated for physical illnesses of the kinds indicated in Table III. The other 22 patients were under investigation or treatment for a mental illness and were confined to bed because of the nature of their treatment, because they were retarded or restless, or because they were weak as a result of advanced age or poor nutrition. There can be no doubt about the desirability of the traditional type of accommodation (*i.e.* a high ratio of bed-space to day-room and other facilities): 77 patients were bedfast or were only occasionally permitted to sit out of bed, and the sixteen who were partly ambulant were either under investigation or were in an early stage of convalescence after an acute physical illness. All the patients needed some supervision because of their mental state, and all but thirteen required limited or close supervision.

The patients in Group A also had in common their needs for medical and nursing care. They all required frequent medical attention and skilled nursing, and 76 needed basic nursing as well (20 were incontinent). But because the patients required mental supervision, the wards would have to be staffed mainly, and perhaps entirely, by nurses with experience of mental as well as physical illness.

Even if it is agreed that the 93 patients in Group A needed traditional ward accommodation with supervision, it may be asked whether it would be desirable to have in the same unit patients being treated for physical and for mental illnesses. The answer to this question must turn largely upon the numbers for whom it is possible to plan (the 93 were drawn from a population of over a million) and upon whether the accommodation is to be designed specifically for the purpose, or adapted from existing facilities. Evidently if new accommodation is designed for a relatively large number of patients with most of their needs in common, it is possible to have regard for lesser differences in requirements which could scarcely be considered in a smaller or in an existing unit. But it seems to us that the overriding considerations are that all these patients needed traditional ward units (this is not of course to pass judgement on the optimum size or type of ward), and all needed skilled nursing, which, because of their mental state, would have to be undertaken mainly or entirely by mental nurses. The fact that some patients needed the services of a general physician, others of a surgeon, and still others of a psychiatrist, seems less significant from the point of view of their location, so long as all the patients are in a hospital which can provide the requisite services. For they all required the general supervision of psychiatric staff, and the numbers attended by any other specialist were probably not large enough to make it practicable to create a separate unit. Nevertheless some subdivision might be desirable within the wards: for example the segregation of patients with tuberculosis who need barrier nursing, and of those whose mental condition makes it necessary for them to be closely supervised. These reservations to not affect our main conclusion, that from the point of view of their accommodation and staff requirements these 93 patients formed a unit, and had more in common with one another than with any other patients in the mental hospitals.

Ward Accommodation without Supervision (Group B).—It was suggested that eight patients (of 459) needed traditional ward accommodation without supervision. They were being treated for physical illnesses, of which three involved the central nervous system (epilepsy, syphilis, paraplegia) and one each of the following: pulmonary tuberculosis, antepartum haemorrhage, thyroid adenoma, diarrhoea of unknown origin, and fracture of femur. It was concluded that they needed traditional ward accommodation, because six patients were either bedfast, or only occasionally permitted to sit out of bed, and the two who were ambulant were under investigation for epilepsy and general paralysis of the insane. It is evident that their requirements were medical treatment and nursing care: all needed skilled nursing and five needed basic nursing as well. In view of the fact that they had no mental symptoms severe enough to require supervision, there could be no possible objection to employment of nurses with a general training. The most satisfactory way in which to provide for them would be to admit them to the appropriate wards of general hospitals.

Closed Hostel-Type Accommodation (Group C).—There were 358 ambulant patients who needed full hospital services because they were under investigation or active treatment of a mental illness. Because they are ambulant such patients do not require traditional ward accommodation, but could be cared for in facilities of the hostel type with a high ratio of day-room and other amenities to bed-space. In Table XI they have been subdivided according to the degree of supervision: 117 would have to be restricted to their own block, whereas the remainder (241) could move freely within the hospital community. It may be questioned whether in practice it would be possible to separate patients in these two groups, all of whom require some supervision. But it would probably be easier to staff the units if patients needing a considerable amount of personal supervision were separated from those who needed

little or no supervision so long as they remained within the hospital grounds.

Of the 117 patients suggested for closed hostel-type accommodation, eight needed close supervision because they were restless, confused, or aggressive, and the remainder could not be allowed unaccompanied outside their unit. Because of the nature of their medical treatment they all required skilled mental nursing. Although they were ambulant, 20 per cent. needed basic nursing, such as feeding, washing, and dressing, and in the case of incontinent patients attention to bladder and bowel.

These patients are also distinguished from those best accommodated in traditional wards (Groups A and B) by their need for some sort of occupation. The question of productive work does not of course arise in respect of patients undergoing investigation or treatment, but it was estimated that about 70 per cent. could benefit from occupational therapy. A considerable proportion (about a third) of those who might have benefited were not receiving this service, which was more deficient than any other considered in the inquiry.

Open Hostel-Type Accommodation (Group D).—Although they were also undergoing intensive investigation and treatment because of their mental illness, the remaining 241 patients were able to move without supervision within the hospital. Again because of their treatment they all needed skilled nursing, but only twelve needed basic nursing (Table XII). The majority of them (88 per cent.) required occupational therapy, which in most cases was being provided. The difference from the previous group is due to the fact that these patients were able to leave the ward and go to the occupational therapy centre.

As compared with Group C patients, those suggested for an open block were much less disturbed and had more insight into the nature of their illness. It seems very undesirable that they should share accommodation with patients who are grossly disturbed. They had much in common with those now treated in some hospitals as out-patients, and with the non-statutory patients accepted as in-patients by early treatment centres.

(*d*) Summary

459 patients (13 per cent. of the total mental hospital population of 3,555) were undergoing investigation or treatment of a mental or physical illness which made it necessary for them to have access to the full resources of a hospital. Those being treated for a mental illness (83 per cent.) were almost all continent and fully ambulant; they all needed skilled nursing, and 13 per cent. needed basic nursing (washing, dressing, feeding, lifting, attention to bladder and bowels, etc.) as well. Patients treated for a physical illness (17 per cent.) were affected by a variety of medical and surgical conditions (pulmonary tuberculosis, cardiovascular disease, traumatic and orthopaedic conditions, etc.). Most of them were confined to bed and many were incontinent. They all required skilled nursing and 85 per cent. required basic nursing. All the patients treated for a mental illness needed some supervision, but two-thirds were restricted only insofar as they were not allowed outside the hospital grounds. Nine-tenths of those with a physical illness required supervision.

It is suggested that the two considerations which have most bearing on the type of accommodation and staff required for patients needing full hospital facilities are their mobility and the supervision they require. Accordingly the data were used to divide the patients into four groups:

(A) 93 patients (20 per cent. of 459) were bedfast or only partly ambulant, and required traditional ward accommodation with supervision because of their mental state. They all needed to be under the general supervision of psychiatrists, but 71 also needed the services of general medical and surgical staff because they were physically ill. In view of their mental condition they had to be attended by nurses with experience of mental as well as physical illness.

Table XII

BASIC NURSING NEEDED BY PATIENTS WHO REQUIRED FULL HOSPITAL FACILITIES AND HOSTEL-TYPE ACCOMMODATION

Accommodation	Basic Nursing								All Patients	
	Constant		Regular		Occasional		None			
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Closed Block (Group C)	4	3·4	19	16·2	1	0·8	93	79·5	117	100
Open Block (Group D)	1	0·4	8	3·3	3	1·2	229	95·0	241	100

(B) Eight patients were bedfast or only partly ambulant, and required traditional ward accommodation without supervision. These patients were under treatment for physical illnesses without mental complications, and could have been cared for by the medical and nursing staff of a general hospital.

(C) 117 ambulant patients needed hostel-type accommodation (a high ratio of day-room and other amenities to bed-space) with restriction to their own block because of their mental state. They were under active investigation or treatment of mental illness (without physical complications), and needed to be under the care of psychiatrists assisted by nurses with experience of mental illness. Most of them needed occupational therapy.

(D) 241 ambulant patients needed hostel-type accommodation, but did not require supervision so long as they remained within the hospital. In other respects their needs were the same as those of patients in Group C.

II. Patients Needing Limited Hospital Facilities

Much the largest of the three classes into which Birmingham residents in mental hospitals were divided consisted of 2,655 patients (75 per cent. of the total) thought to need limited hospital facilities. Limited hospital facilities include such simple services as basic nursing, supervision of mentally-ill patients, and occupational training, but not the more comprehensive services—investigation, intensive medical treatment, skilled nursing—which can be provided only where the full range of hospital services is available. Most of the patients needing limited facilities were either confined to bed by chronic physical illness, or were ambulant but in need of supervision because of their mental state. In respect of requirements they were therefore distinguishable both from the patients referred to in the previous section who needed full hospital services, and from those—discussed in the following section—who were considered suitable for hostel accommodation without mental supervision.

(*a*) Type of Patient

Table XIII gives the distribution of the 2,655 patients according to the diagnosis of their mental illness. The distribution is very similar to that of the whole mental hospital population (Garratt and others, 1957: Table V)—almost inevitably, since they comprise about three-quarters of it. The only obvious difference is the greater excess of females. This is due largely to the sex ratio of patients with manic depressive reaction, paranoid state, senile psychosis, and involutional melancholia, among whom the relative numbers of females to males were 2 to 1, 3 to 1, 5 to 1, and 10 to 1 respectively. A similar excess of females was recorded in a previous investigation of chronic patients in a Birmingham mental hospital (Cross and others, 1957). It is evidently much too great to be attributable wholly to differences between the numbers of males and females in the related age groups of the general population.

TABLE XIII

DIAGNOSIS OF PATIENTS WHO REQUIRED LIMITED HOSPITAL FACILITIES

Diagnosis*	Males	Females	All Patients	
			Number	Per cent.
Schizophrenia (300) ..	508	640	1,148	43·2
Manic Depressive Reaction (301)	55	122	177	6·7
Involutional Melancholia (302)	7	78	85	3·2
Paranoid State (303) ..	67	197	264	9·9
Senile Psychosis (304) ..	69	353	422	15·9
Other Psychoses (305–309)	66	103	169	6·4
Psychoneuroses (310–18) ..	9	13	22	0·8
Disorders of Character and Behaviour (320–24; 326)	9	14	23	0·9
Mental Defect (325) ..	57	94	151	5·7
Syphilis (025)	26	18	44	1·7
Epilepsy (353)	36	52	88	3·3
Other Disorders	35	27	62	2·3
Total	944	1,711	2,655	100

* Numbers in brackets are list numbers used in 1948 International Statistical Classification.

It seems unnecessary to examine in detail the age distribution and duration of stay which, for the reason previously given, inevitably resembled those of the whole hospital population. But, since most patients needing limited facilities were chronic patients, they included a greater proportion of older patients and of patients who had been in hospital for long periods.

For the purpose of assessing their requirements, patients in this class were divided according to whether their needs were due primarily to a mental or a physical illness (Table XIV). Although it is not a precise guide, the distinction is a useful one. Most of the mentally ill were ambulant and needed supervision and occupational training, but little nursing. Most of the physically ill were bedfast and needed basic nursing; occupational training was of course relatively less important.

TABLE XIV

TYPE OF ILLNESS FOR WHICH PATIENTS REQUIRED LIMITED HOSPITAL FACILITIES

Patients Needing Limited Hospital Facilities for:	Males		Females		All Patients	
	No.	Per cent.	No.	Per cent.	No.	Per cent.
Mental Illness ..	846	89·6	1,463	85·5	2,309	87·0
Physical Illness ..	98	10·4	248	14·5	346	13·0
All Patients	944	100	1,711	100	2,655	100

The distribution according to diagnosis of the 2,309 patients whose need of hospital facilities was attributed mainly to mental illness does not differ in any important respect from that of all patients in this class (Table XIII). On the other hand, the 346 patients whose needs were attributable mainly to physical illness were fairly sharply distinguished in respect of the nature of their mental illness. The proportion of schizophrenics (8·4 per cent.) was much lower and that of senile (48·8 per cent.) and other psychotics (12·4 per cent.) much higher. The physical illnesses upon which their need for mental hospital care largely rested are given in Table XV. More than half of the patients had no more specific diagnosis than infirmity due to old age and most of the others were affected by degenerative conditions. These observations merely reflect the fact that the physically ill consisted mainly of senile patients, most of whom also had mental symptoms.

TABLE XV

TYPE OF PHYSICAL ILLNESS FOR WHICH PATIENTS REQUIRED LIMITED HOSPITAL FACILITIES

Type of Illness	Males	Females	All Patients	
			Number	Per cent.
Infirmity of Old Age ..	42	148	190	54·9
Organic Disease of Central Nervous System ..	22	31	53	15·3
Diseases of Heart and Circulation	13	7	20	5·8
Arthritis (all forms) ..	3	15	18	5·2
Cerebro-Vascular Disease	5	12	17	4·9
Chronic Bronchitis ..	5	5	10	2·9
Pulmonary Tuberculosis ..	—	10	10	2·9
Malignant Disease ..	2	7	9	2·6
All Other Conditions ..	6	13	19	5·5
Total	98	248	346	100

(*b*) PATIENTS' NEEDS

Medical Treatment.—The reader is reminded that patients who needed the sort of medical attention which could be given only where the full range of hospital facilities was available have, by definition, been excluded from this class. Treatment of those without associated physical illness consisted of administration of tranquillizers (43 per cent.) or other sedatives (1 per cent.); more than half were receiving no specific treatment. (Once again it should be said that, as the psychiatrists were in a position to give the treatment which they considered necessary, the treatment required is the same as that actually received.) However, it is generally agreed that drug therapy is a less important part of the medical care of this class of patient than occupational training, which is discussed below. The patients with a physical illness (Table XV) needed only the occasional medical supervision required by the chronic sick whose illnesses are no longer under active investigation or treatment. Of course they have acute exacerbations from time to time when they require frequent medical attention (and skilled nursing). But, for the purposes of the present analysis, a patient in this stage of illness (for example with pneumonia or heart failure) would be included among those considered to need full hospital facilities. It is natural to ask why these physically ill patients were in mental hospitals: 57·8 per cent. of them were physically ill when they were admitted and were accepted in a mental hospital because of associated mental symptoms; and 42·2 per cent. had developed their chronic physical illness since their admission (Table XVI).

TABLE XVI

LOCATION OF PATIENTS WHO REQUIRED LIMITED HOSPITAL FACILITIES FOR PHYSICAL ILLNESS AT THE ONSET OF THAT ILLNESS

Location	Home		Mental Hospital	Other*	Total
	Direct Admission	Transfer from General or Chronic Sick Hospital			
Number	181	12	146	7	346
Per cent.	52·3	3·5	42·2	2·0	100

* Municipal or voluntary home for elderly or blind.

Nursing Care.—It has been said previously that nursing care was one of the most significant considerations in assigning patients to the three major classes. Patients needing full hospital facilities all required skilled nursing; patients needing limited hospital facilities required no skilled nursing but

TABLE XVII

BASIC NURSING NEEDED BY PATIENTS WHO REQUIRED LIMITED HOSPITAL FACILITIES

Limited Hospital Facilities Required for:	Constant		Regular		Occasional		None		All Patients	
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Mental Illness ..	51	2·2	392	17·0	35	1·5	1,831	79·3	2,309	100
Physical Illness ..	150	43·4	182	52·6	14	4·1	—	—	346	100

many of them required basic nursing; patients needing no hospital facilities and considered suitable for hostel accommodation required no nursing. Evidently the nursing care would provide a precise index, were it not for inclusion in the second class (limited hospital facilities) of some patients who did not need basic nursing, but required supervision because of their mental state.

The nursing requirements of patients in this class are shown in Table XVII. All of those with a physical illness needed the simple personal services—washing, dressing, feeding, etc.—required by the chronic sick. More than half of them were incontinent (Table XVIII), and about two-thirds were bedfast (Table XIX). Of the mentally-ill patients on the other hand, only about one-fifth needed basic nursing (Table XVII), and this is understandable, since most of them were continent (Table XVIII) and fully ambulant (Table XIX). This is of course the source of the only inconsistency in the use of nursing as an index in classifying patients. It was considered right to include among those suitable for limited hospital facilities mentally ill patients who made few demands on the nursing services in any respect other than their need for supervision and training. Unless they were assigned to a separate class, it seemed preferable to group these patients with those with a physical illness who needed basic nursing, rather than with the patients considered suitable for hostel facilities who were wholly independent of the nursing services.

TABLE XVIII

CONTINENCE OF PATIENTS WHO REQUIRED LIMITED HOSPITAL FACILITIES

Limited Hospital Facilities Required for:	Incontinent				Continent		All Patients	
	Wholly		Partly					
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Mental Illness	209	9·1	63	2·7	2,037	88·2	2,309	100
Physical Illness	190	54·9	7	2·0	149	43·1	346	100

Supervision.—In Table XX patients are distributed according to the amount of supervision they required. All those with a mental illness needed some supervision, although more than half of them were permitted to move freely within the hospital grounds and less than 1 per cent. needed close personal supervision. Most of the physically ill patients also had to be supervised, but 59 of them

TABLE XIX

MOBILITY OF PATIENTS WHO REQUIRED LIMITED HOSPITAL FACILITIES

Limited Hospital Facilities Required for:	Bedfast		Sitting Out of Bed		Ambulant				All Patients	
					Partly		Fully			
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Mental Illness ..	9	0·4	8	0·3	117	5·1	2,175	94·2	2,309	100
Physical Illness ..	219	63·3	102	29·5	24	6·9	1*	0·3	346	100

* Congenital syphilis with epilepsy, too many fits for hostel.

TABLE XX

SUPERVISION NEEDED BY PATIENTS WHO REQUIRED LIMITED HOSPITAL FACILITIES

Limited Hospital Facilities Required for:	Supervision								All Patients	
	Close		Limited		Open		None			
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Mental Illness ..	15	0·6	1,058	45·8	1,236	53·5	—	—	2,309	100
Physical Illness ..	11	3·2	276	79·8	—	—	59	17·1	346	100

needed no supervision of any kind. Since these patients were confined to bed the question of leaving the hospital and its grounds unaccompanied did not arise.

Occupation.—Although patients in this class needed little specific medical therapy, many of them required training, of which a suitable occupation is the most tangible feature. When they are found suitable work, patients with chronic mental illness often improve, sometimes to such an extent that they become fit to return home and take up normal employment.

Most of the 346 patients with physical illness were bedfast and consequently their occupational needs were limited. 52 (15 per cent.) of them required occupational therapy in the wards (only twenty were receiving it); but the remaining 294 were so grossly disabled that any form of occupation was precluded (Table XXI).

Almost all the 2,309 patients who required limited hospital facilities because of mental illness were ambulant (Table XIX), and their occupation was therefore a matter of considerable importance. About one-quarter were capable of productive work and nearly one-third required occupational therapy (Table XXI). The remainder were thought to be unsuitable for occupational training, being too inaccessible, retarded, or degraded. Some of these patients were encouraged to perform simple stereotyped tasks in the ward, but the majority were completely unoccupied and appeared to be incapable of doing anything.

(*c*) Accommodation Required

It has already been suggested that the two considerations which have most bearing on the type of accommodation and staff required for the care of the mentally ill are the amount of supervision they require and their mobility; the 2,655 patients needing limited hospital facilities have been classified accordingly (Table XXII). We have also distinguished patients according to whether their needs were attributable mainly to mental or to physical illness. Again, and for reasons considered previously, the criteria of mobility and supervision are not a precise guide to the facilities and staff required. The physically ill were not all bedfast patients who were unsupervised, and the mentally ill were not all ambulant patients who were supervised. But by grouping the cells of Table XXII we have obtained four groups of patients identified according to these two features of care (Table XXIII, overleaf).

Group A. Bedfast or only partly ambulant patients, all of whom required supervision.

Group B. Bedfast or only partly ambulant patients who required no supervision.

Group C. Ambulant patients restricted within the hospital community.

Table XXI

OCCUPATION NEEDED BY PATIENTS WHO REQUIRED LIMITED HOSPITAL FACILITIES

Limited Hospital Facilities Required for:	None		Light Work on Ward		Occupational Therapy		Productive Work		Total	
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Mental Illness ..	677	29·3	337	14·6	698	30·2	597	25·9	2,309	100
Physical Illness ..	294	85·0	—	—	52	15·0	—	—	346	100

Table XXII

CLASSIFICATION OF PATIENTS WHO REQUIRED LIMITED HOSPITAL FACILITIES

Patients		Bedfast or Sitting Out of Bed		Partly or Fully Ambulant		All Patients
Illness		Mental Only	Physical	Physical	Mental Only	
Supervision ..	Close	(A) 1	9	2	(C) 14	26
	Limited	7	264	12	1,051	1,334
	Open	9	—	—	(D) 1,227	1,236
	None	—	(B) 48	11	—	59
Total		17	321	25	2,292	2,655

Group D. Ambulant patients unrestricted within the hospital community but not allowed outside the hospital grounds.

Although this classification is the same as that employed in the previous section, the patients considered there were of course distinguished sharply by their need for frequent medical and skilled nursing care.

It is suggested that these four groups of patients could have been cared for most conveniently in different units, which are discussed separately below. For the purpose of this classification six patients who were only temporarily bedfast (three with coryza, two with septic feet, and one with a fractured arm) were transferred from Group A to Group C (one patient) and Group D (five patients). This explains the small difference between numbers in Tables XXII and XXIII.

TABLE XXIII

TYPE OF ACCOMMODATION NEEDED BY PATIENTS WHO REQUIRED LIMITED HOSPITAL FACILITIES

Accommodation	Traditional Ward	Hostel-Type	Total
Closed Block	(A) 298*	(C) 1,066*	1,364
Open Block	(B) 59	(D) 1,232*	1,291
Total	357	2,298	2,655

* Small differences between numbers in Table XXII and XXIII are accounted for in the text.

Ward Accommodation with Supervision (Group A).—It was thought that 298 patients (11 per cent. of 2,655) should be cared for in traditional wards which provided some personal supervision. 287 were physically ill, and their need for traditional accommodation (a high ratio of bed space to day-room and other facilities) is evident from the fact that almost all of them were confined to bed. For the fourteen physically ill patients who were partly ambulant (Table XXII), it would probably be desirable to provide a small day room, but their movements were so restricted that it would still be preferable to care for them in a traditional ward. The remaining eleven in Group A (after transfer of six to Groups C and D) were catatonic or confused patients confined to bed because of their mental state.

The staff required in such a unit is readily stated. The patients all needed simple personal services, but, by present standards at least, they required little medical attention or skilled nursing over long periods. They therefore did not need immediate access to the full resources of a hospital, but required only general medical supervision of the kind commonly provided by a general practitioner for patients in their own homes. The imperative demand was for basic nursing, which, if adequately supervised, did not need to be given by skilled staff. But as the patients all had abnormal mental symptoms, they would probably be considered unsuitable for care by nurses in training for the general register. Because they were restricted to bed, and many of them were unlikely ever to be ambulant again, occupational facilities were far less important than in the case of the ambulant patients in Groups C and D.

We shall not attempt to consider in great detail the needs of patients or the design of the unit. But again it is worth noting that there were twelve patients who required close supervision because they were severely disturbed (mainly noisy and confused senile psychotics, and four aggressive schizophrenics) and nine who had chronic pulmonary tuberculosis. This suggests that, if a unit were designed for the purpose and on a sufficient scale, it would be desirable to effect some segregation within it. But administratively the differences seem to us less significant than the features which all the patients in Group A had in common: their need for traditional ward-type accommodation with supervision because of their mental state.

Ward Accommodation without Supervision (Group B).—59 patients (2 per cent. of 2,655) needed traditional ward accommodation in which it was unnecessary to provide supervision. They were physically ill patients with mental symptoms of so trivial a kind that they had no bearing on their care; 27 had organic diseases of the central nervous system, fifteen had other organic diseases, and the remaining seventeen had no more specific diagnosis than infirmity associated with old age. They needed traditional accommodation because 48 were bedfast or occasionally sat out of bed, and the other eleven were only partly ambulant.

Like those in the previous group these patients required only general medical supervision and basic nursing. They differed sharply, however, in having no mental symptoms which needed supervision, and, if they had not been restrained by infirmity or physical illness, there could have been no objection to their leaving hospital unaccompanied. In terms of needs they were evidently identical with patients cared for in hospitals for the chronic sick, and that is of course where those responsible for administration of mental hospitals would prefer to see them. Again, because of their very limited movement, the question of occupation was of secondary importance.

Closed Hostel-type Accommodation (Group C).—2,298 (86 per cent. of the 2,655 patients needing limited hospital facilities) were fully ambulant, and were therefore considered suitable for hostel-type accommodation. These patients comprise the hard core of the mental hospital population. They were affected by mental illness without an associated physical illness. About half of them were schizophrenics, unlike the patients with an associated physical illness (Groups A and B) of whom half were senile psychotics. All of them required some supervision because of their mental state, but they were divided into two groups according to whether they were or were not confined to the wards (Groups C and D respectively). Once again it might be asked whether in practice provision of separate accommodation would be feasible, and it would perhaps be less essential than the parallel separation in the case of patients needing full hospital facilities (which would more sharply divide those with insight from disturbed patients). Nevertheless it seems to us that from the point of view of the patients it would be desirable, and administratively it would be at least convenient, to separate those who could be permitted to leave their residential blocks unaccompanied, from those who could not.

There were 1,066 patients who needed a degree of personal supervision which made it undesirable for them to leave the wards unaccompanied; fourteen of them required close supervision. They needed little medical attention, but 30 per cent. of them required basic nursing (Table XXIV). One of their most important demands was suitable employment; 14·4 per cent. were capable of productive work, that is of work which made a contribution to the hospital community, and only 38·2 per cent. were thought unsuitable for any type of occupation (Table XXV). As a further indication of the condition of patients it may be noted that about 70 per cent. were considered unsuitable for parole. In practice, 90 per cent. never left the hospital.

Open Hostel-type Accommodation (Group D).—The last group consists of the 1,232 ambulant patients who were allowed to move freely within the hospital community, but were considered unsuitable to leave the grounds alone. Their medical and nursing needs were approximately the same as those of the previous group: occasional medical supervision with basic nursing in about 10 per cent. of cases (Table XXIV). Only one-third of them were judged unsuitable to be released on accompanied parole, and in practice about one-quarter were receiving it.

Perhaps the most interesting feature of the care of this group of patients is their employment (Table XXV). Only about one-fifth were incapable of any occupation, and about one-third could undertake productive work. Patients of this type do in fact make a considerable contribution to the work of mental hospitals, and if they were not available it would be necessary to bring in workers to replace them.

TABLE XXIV

BASIC NURSING NEEDED BY PATIENTS WHO REQUIRED LIMITED HOSPITAL FACILITIES AND HOSTEL-TYPE ACCOMMODATION

Accommodation	Basic Nursing								All Patients	
	Constant		Regular		Occasional		None			
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Closed Block (Group C)	34	3·2	279	26·2	7	0·7	746	70·0	1,066	100
Open Block (Group D)	14	1·1	105	8·5	28	2·3	1,085	88·1	1,232	100

TABLE XXV

OCCUPATION NEEDED BY PATIENTS WHO REQUIRED LIMITED HOSPITAL FACILITIES AND HOSTEL-TYPE ACCOMMODATION

Accommodation	Occupation Needed								All Patients	
	None		Light Work on Ward		Occupational Therapy		Productive Work			
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Closed Block (Group C)	407	38·2	157	14·7	348	32·6	154	14·4	1,066	100
Open Block (Group D)	259	21·0	180	14·6	350	28·4	443	36·0	1,232	100

Table XXVI shows the kind of work actually done by those patients in Groups C and D who were engaged on productive work.

TABLE XXVI

PRODUCTIVE WORK DONE BY PATIENTS WHO REQUIRED LIMITED HOSPITAL FACILITIES AND HOSTEL-TYPE ACCOMMODATION

Occupation	Males	Females	All Patients	
			Number	Per cent.
Ward Cleaner	98	157	255	45·2
Kitchen or Canteen Assistant	31	56	87	15·4
Garden or Farm Labourer	70	—	70	12·4
Sewing Room Assistant ..	—	62	62	11·0
Laundry Worker	—	35	35	6·2
Other	46	9	55	9·8
Total	245	319	564	100

(*d*) SUMMARY

2,655 patients (75 per cent. of the total mental hospital population of 3,555) required no skilled nursing and only occasional medical attention, but needed basic nursing or supervision or both (referred to as limited hospital services). 87 per cent. had a mental illness without physical complications: most of them were ambulant and needed supervision and occupational training but little nursing. About one-quarter of them were capable of productive work. 13 per cent. were physically ill (more than half were elderly senile patients): most of them were bedfast and needed both basic nursing and supervision. Patients were again divided into four groups according to their mobility and supervision:

(A) 298 patients were bedfast or only partly ambulant, and required traditional ward accommodation with supervision. They needed only occasional medical attention, of the kind commonly given by general practitioners to patients in their own homes. Their essential requirement was basic nursing, provided in view of their mental state, by nurses with experience of mental illness.

(B) 59 patients were bedfast or only partly ambulant, and required traditional ward accommodation without supervision. Like the previous group they needed basic nursing but only occasional medical attention. Because they did not require supervision they could be cared for by nurses without experience of mental illness. In terms of the nature of their illness and their needs these patients are identical with those cared for in hospitals for the chronic sick.

(C) 1,066 ambulant patients needed hostel-type accommodation with restriction to their own block because of their mental state. They needed occasional medical attention with (in 30 per cent. of cases) basic nursing by nurses with experience of mental illness. Perhaps their most important requirement was training and employment. About 15 per cent. were capable of productive work.

(D) 1,232 ambulant patients needed hostel-type accommodation without supervision so long as they remained within the hospital. In other respects their needs were the same as those of patients in Group C, although the proportion capable of productive work (about one-third) was higher.

III. PATIENTS NOT REQUIRING HOSPITAL FACILITIES

After exclusion of the previous two classes, we are left with 441 patients (12 per cent. of 3,555 Birmingham patients in mental hospitals) who in the opinion of the psychiatrists required none of the facilities usually associated with a hospital. They did not need investigation, specific medical treatment, or skilled nursing (services demanding full hospital facilities), and they had neither physical disability serious enough to require basic nursing nor mental abnormality for which supervision was necessary (services demanding limited hospital facilities). Their circumstances were analogous to those of the patients in hospitals for the chronic sick who do not need specific medical or nursing care but remain in hospital chiefly for social reasons. It might be thought that all such patients in the mental hospitals could have been discharged if they had had a suitable home. This was not the case. Although none of them required traditional hospital services, more than half were considered to be unable to live wholly independent lives. They needed no personal supervision but they did need to live in an organized community which could provide assistance with such matters as training and employment. In the discussion which follows, the 441 patients are divided into two groups according to whether or not, in the opinion of the psychiatrist, they could have been discharged if a suitable home had been available.

(*a*) TYPE OF PATIENT

Table XXVII (opposite) shows the distribution of the 441 patients by the diagnosis of their mental illness. As compared with the whole mental hospital population (Garratt and others, 1957: Table V), there were fewer schizophrenics and senile psychotics, and more manic depressives, mental defectives, epileptics, and individuals with disorders of

TABLE XXVII

DIAGNOSIS OF PATIENTS WHO DID NOT REQUIRE HOSPITAL FACILITIES

Diagnosis*	Males	Females	All Patients	
			Number	Per cent.
Schizophrenia (300) ..	49	54	103	23·4
Manic Depressive Reaction (301)	18	46	64	14·5
Involutional Melancholia (302)	2	21	23	5·2
Paranoid State (303) ..	17	26	43	9·8
Senile Psychosis (304) ..	7	27	34	7·7
Other Psychoses (305–09)	11	10	21	4·8
Psychoneuroses (310–18) ..	5	8	13	2·9
Disorders of Character and Behaviour (320–24; 326)	18	21	39	8·8
Mental Defect (325) ..	17	27	44	10·0
Syphilis (025)	1	4	5	1·1
Epilepsy (353)	16	16	32	7·3
Other Disorders	15	5	20	4·5
Total	176	265	441	100

* Numbers in brackets are list numbers used in 1948 International Statistical Classification.

character and behaviour. Once again, there was an excess of females, which was particularly marked for certain kinds of illness (involutional melancholia, manic depressive reaction, and senile psychosis). The 441 patients were a little older (two-fifths were over 65) than the total hospital population, and had been in a mental hospital for about the same length of time (nearly two-thirds for more than 5 years). A third of the patients were still technically certified, although not in need of supervision. In part this anomaly is accounted for by the fact that after 7 years' residence certification orders are reviewed only at 5-yearly intervals, unless the question of discharge from hospital arises. We may summarize these observations by stating that, in respect of the diagnosis of their mental illness, their age, and the duration of stay in hospital, the patients considered not to need hospital facilities were fairly representative of the whole mental hospital population. For obvious reasons they differed in their status under the Lunacy and Mental Treatment Acts, the surprising feature being that a considerable proportion of them were still certified.

We have no information about the circumstances in which these patients who no longer needed hospital care were admitted to hospital. Some no doubt were more seriously ill than at the time of our assessment, but their illnesses had remitted or responded to treatment; others may have been admitted chiefly for social reasons. It was considered impossible to assess the relative importance of these two influences at the time of our survey, which in most cases was many years after the original admission.

Inquiry why the patients had not been discharged elicited the distinction already referred to between those who needed to live in an organized community (54·9 per cent.) and those who could have been released to a suitable home (Table XXVIII). Of the 179 patients who could have been discharged, 74 had no home, and 105 could not return to their homes because relatives or friends were unable or unwilling to receive them. Twenty patients were waiting to be discharged at the time of investigation.

TABLE XXVIII

REASON WHY PATIENTS WHO DID NOT REQUIRE HOSPITAL FACILITIES HAD NOT BEEN DISCHARGED

Reason		Males	Females	All Patients	
				Number	Per cent.
Needed Life in Organized Community		119	123	242	54·9
Home Unsuitable	Relatives or Friends Incapable	13	47	60	13·6
	Relatives or Friends Unwilling	12	33	45	10·2
	No Home	25	49	74	16·8
Awaiting Discharge ..		7	13	20	4·5
Total		176	265	441	100

Since the disposal of the patients who did not require hospital accommodation is a matter of considerable practical importance, it was thought necessary to inquire whether they had any physical or mental symptoms which might prejudice the possible arrangements for their care. Although they were all ambulant, continent, and able to feed, wash, and dress themselves, about one-quarter had physical disabilities (Table XXIX, overleaf), and needed a little assistance from time to time. (For example, 29 of the 31 epileptics, although well controlled, had occasional fits, and the mobility of 27 other patients was limited to some extent by disorders such as Parkinsonism, club-foot, and old hemiplegia). And although none of the patients had a mental abnormality serious enough to require supervision, about half of them still had abnormal

TABLE XXIX

PHYSICAL DISABILITY IN PATIENTS WHO DID NOT REQUIRE HOSPITAL FACILITIES

Physical Disability	Accommodation Needed				All Patients	
	Hostel in Organized Community		Home Only			
	No.	Per cent.	No.	Per cent.	No.	Per cent.
Epileptic Fits ..	14	5·8	15	7·5	29	6·6
Disorders affecting Locomotion	10	4·1	17	8·5	27	6·1
Deaf-mutism ..	9	3·7	4	2·0	13	2·9
Partial-sight ..	5	2·1	7	3·5	12	2·7
Other..	30	12·4	13	6·5	43	9·7
None	174	71·9	143	71·8	317	71·9
Total	242	100	199	100	441	100

mental symptoms (Table XXX) and some occasionally needed assistance or guidance.

TABLE XXX

ABNORMAL MENTAL SYMPTOMS OR BEHAVIOUR IN PATIENTS WHO DID NOT REQUIRE HOSPITAL FACILITIES

Abnormal Mental Symptoms or Behaviour	Accommodation Needed				All Patients	
	Hostel in Organized Community		Home Only			
	No.	Per cent.	No.	Per cent.	No.	Per cent.
Simple, Immature ..	40	16·5	21	10·6	61	13·8
Residual Delusions ..	31	12·8	24	12·1	55	12·5
Retarded, Apathetic	21	8·7	8	4·0	29	6·6
Mild Depression ..	15	6·2	9	4·5	24	5·4
Other..	39	16·1	16	8·0	55	12·5
None	96	39·7	121	60·8	217	49·2
Total	242	100	199	100	441	100

(*b*) PATIENTS' NEEDS

Medical Treatment and Nursing Care.—It has already been said that none of these patients was considered to require investigation, or the kind of medical treatment which would have made it necessary to be in hospital. About a third were receiving tranquillizers or other drug therapy (sodium amytal, amphetamine, etc.), but this treatment is commonly given to patients in their own homes and had no bearing on the question of disposal. The reader is also reminded that no patient was considered to be independent of hospital facilities (*i.e.* was included among the 441 under consideration) if he needed any form of nursing care or personal supervision.

Occupation.—Perhaps the most important facet of the care of this group of patients is their occupation. Their needs are assessed in Table XXXI. 267 (about three-fifths) were capable of productive work, usually under sheltered conditions, and a further 33 could have been employed after a period of rehabilitation. It is perhaps surprising that the proportion who could do productive work was higher among patients who needed to live in a sheltered environment, than among those who could have been discharged to a suitable home. This anomaly is explained by the fact that the latter group contained a considerably greater proportion of elderly patients near or beyond retirement age. About 10 per cent. needed neither productive work nor occupational therapy, but this was due to age rather than mental abnormality.

TABLE XXXI

OCCUPATION NEEDED BY PATIENTS WHO DID NOT REQUIRE HOSPITAL FACILITIES

Occupation Needed	Accommodation Needed				All Patients	
	Hostel in Organized Community		Home Only			
	No.	Per cent.	No.	Per cent.	No.	Per cent.
None	17	7·1	27	13·5	44	9·9
Occupational Therapy	33	13·6	64	32·2	97	22·0
Occupational Rehabilitation	24	9·9	9	4·5	33	7·5
Sheltered Productive Work	155	64·0	86	43·2	241	54·6
Open Productive Work	13	5·4	13	6·5	26	5·9
Total	242	100	199	100	441	100

The work which patients were actually doing is shown in Table XXXII (opposite). Most of those capable of productive work (243 of 267) were employed on it, but a considerable number who were not receiving occupational rehabilitation (28) could have benefited from it. About one-quarter of the patients had no occupation other than occasional light work on the ward.

Parole and Visiting.—The extent to which patients were leaving hospital on parole is assessed in Table XXXIII. Nearly half were not receiving parole: because the possibility appeared not to have been considered (79 patients); because the patients were uninterested (65); because they were judged unsuitable to leave hospital alone (43) and had no relative

TABLE XXXII

PRESENT OCCUPATION OF PATIENTS WHO DID NOT REQUIRE HOSPITAL FACILITIES

Present Occupation	Accommodation Needed				All Patients	
	Hostel in Organized Community		Home Only			
	No.	Per cent.	No.	Per cent.	No.	Per cent.
None	46	19·0	71	35·7	117	26·5
Occupational Therapy	30	12·4	46	23·1	76	17·2
Occupational Rehabilitation	3	1·2	2	1·0	5	1·1
Sheltered Productive Work	161	66·5	78	39·2	239	54·2
Open Productive Work	2	0·8	2	1·0	4	0·9
Total	242	100	199	100	441	100

TABLE XXXIV

SLEEPING ACCOMMODATION PROVIDED FOR PATIENTS WHO DID NOT REQUIRE HOSPITAL FACILITIES

Sleeping Accommodation		Accommodation Needed				All Patients	
		Hostel in Organized Community		Home Only			
		No.	Per cent.	No.	Per cent.	No.	Per cent.
Single Room ..		15	6·2	2	1·0	17	3·9
Dormitory	With Patients of Same Type	42	17·4	40	20·1	82	18·6
	With Patients of Different Types	185	76·4	157	78·9	342	77·5
Total		242	100	199	100	441	100

or friend to accompany them. This last reason may be thought to be inconsistent with the basis of the original classification, according to which no one was included among the 441 in this class if he required any personal supervision. Of the 43 patients 24 were at advanced ages, nine were deaf mutes, and seven were simple minded. From the point of view of the original classification, some of these were borderline cases. But while they would have required some degree of personal care outside hospital, it seemed inaccurate to say that they needed to be confined to the hospital grounds because of their mental state.

About one-third of the 441 patients were never visited, and one-quarter were visited only occasionally. The proportions were approximately the same for the two sub-classes into which the group was divided.

Sleeping Arrangements.—Since these patients were among the least disturbed in the mental hospitals, and many of them could have lived in the general community, it was thought of interest to examine the sleeping accommodation provided in the hospitals (Table XXXIV). Seventeen were in single rooms and 82 shared dormitory accommodation exclusively with patients of their own class. The great majority (342), however, shared dormitories with patients from the other classes, some of whom were severely disturbed.

(*c*) ACCOMMODATION REQUIRED

It is suggested that the 441 patients under consideration could have been cared for more economically and more effectively if separated from those in each of the two previous classes. Separate accommodation would have been more economical because they did not require the expensive services of a hospital. It would have been more effective because more attention could have been given to their real needs, such as occupation and recreation, and they would be segregated from patients with more severe forms of physical and mental illness.

In the case of the 242 patients who needed to live in an organized community, it seems evident that institutional accommodation was required. In view

TABLE XXXIII

PAROLE RECEIVED BY PATIENTS WHO DID NOT REQUIRE HOSPITAL FACILITIES

Accommodation Needed	Parole Received		Parole Not Received—Reason:						Total	
			Patient not Interested		Patient Unsuitable to Go Alone		No Obvious Reason			
	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.	No.	Per cent.
Hostel in Organized Community ..	147	60·7	40	16·5	17	7·0	38	15·7	242	100
Home Only ..	107	53·8	25	12·6	26	13·1	41	20·6	199	100
All Patients ..	254	57·6	65	14·7	43	9·8	79	17·9	441	100

of the needs we have described a high ratio of day-room and other facilities to bed space was required. The services needed were those usually provided in residential homes, essentially the domestic services, and in addition adequate facilities for recreation, occupational therapy, and training. Those capable of productive work, and they were the majority, could probably have been employed most conveniently in the hostel or in its grounds.

In the case of the 199 patients who needed only a home it is not so obvious that institutional accommodation was necessary. This is the type of patient for whom a policy of boarding out might be applicable, and for some of them it would no doubt be the best solution, perhaps particularly in the case of elderly patients who are no longer able to work. Those who can do productive work are, however, extremely useful in the present mental hospitals, and if they were boarded out alternative labour would have to be found. If satisfactory institutional accommodation were provided there would probably be no objection to retaining many of them. Their requirements were more or less identical with those of the other 242 patients in this class; and there is no reason why they should be separated from them.

The institutional arrangements which could be made would inevitably depend on the number of patients to be catered for at a single centre, and on whether new facilities were designed for the purpose. But it is worth recalling that although the patients were identified according to their needs, they were by no means a homogeneous group. Among them there were several sub-groups, and to the extent that segregation was possible it might be desirable to separate them. For example, there were 31 epileptics, 61 who were "simple" and inactive, thirteen deaf mutes, and a considerable number of elderly patients, of whom 77 were over 75 years of age.

(*d*) Summary

441 patients (12 per cent. of the total mental hospital population of 3,555) required none of the facilities usually associated with a hospital (*viz.*, medical attention, nursing, or supervision). Their main need was suitable employment, more than three-fifths being capable of productive work. Patients were divided into two groups according to whether they could have been discharged to a suitable home (199) or not (242). Those who could not be discharged were considered to need the support of an organized community. Those who could have been discharged either had no home, or could not return home because relatives or friends were unwilling or unable to receive them. It is suggested that patients of this type might be boarded out, but as they are very useful in hospital it may be desirable to retain them with suitable safeguards for their welfare. Accommodation for all of the patients not requiring hospital facilities should be of hostel type, and separate from that of the previous two classes.

Discussion

In the past, hospitals have not been designed with any close regard for the needs of the patients who occupy them. For example, an examination of all patients in a large hospital for the chronic sick (Lowe and McKeown, 1949) suggested that they could be divided fairly sharply into three classes: those who needed full hospital facilities (such as frequent medical attention and skilled nursing); those who needed only limited hospital facilities (essentially basic nursing—washing, dressing, feeding, attention to bladder and bowels, etc.); and those who needed no hospital facilities but were retained for social reasons. The same mixing of patients with entirely different needs was also observed in sanatoria (Lowe and Geddes, 1952), and it seemed probable that it would be found in mental hospitals and, to a lesser extent, in general hospitals. In the investigation described in this and a previous report (Garratt and others, 1957), we have therefore attempted to examine the medical, nursing, and social needs of all Birmingham patients in mental hospitals, with a view to using this information to assess the type, size, staff, and possibly the site of the mental hospitals which should be provided in future. Here we are concerned with only the first three of these four features.

In the first report it was shown that mental hospital patients could be divided readily into the same three broad classes, according to the type of facilities required for their care. We have now attempted a more detailed assessment of the arrangements needed for the patients in each class. For those requiring full or limited hospital facilities, it is suggested that the two most significant considerations are their mobility and the amount of supervision they require. Patients who are bedfast or only partly ambulant are considered to need traditional ward accommodation, but those who are fully ambulant should be in hostel-type accommodation with a much higher ratio of day-room and other facilities to bed space. The significance of supervision is of course that it largely determines the type of nurse who can be employed. Patients who must be personally supervised because of their mental state require the attention of nurses with experience

of mental illness, whereas there could be no reasonable objection to the use of nurses trained or in training for the general register in the case of patients who do not need supervision. The distinction is perhaps somewhat theoretical in present circumstances, since the mental hospitals are usually isolated and all their nurses are recruited on the understanding that they will attend the mentally ill. But it might be of first rate importance if the mental hospitals were brought into closer association with the general hospitals to the extent of sharing a common nursing staff.

The issues are different in respect of patients in the third class, who required none of the services traditionally associated with a hospital. All of them were ambulant and needed hostel-type accommodation, separate from seriously disturbed patients, with adequate arrangements for training and employment. From the point of view of their care perhaps the most important question is whether they should be boarded out (or more accurately, the extent to which they should be boarded out) or retained in hospital, where they make a valuable contribution to essential work.

It is evident, of course, that the results of this analysis reflect the care formerly given to the mentally ill, and can be expected to change both as arrangements at home and in hospital improve, and as new methods of treatment become available. This merely reminds us that no appraisal can be final. Nevertheless, a hospital system based on an assessment of contemporary requirements is less likely to become redundant, and will be more readily adapted to changing needs than one which reflects earlier needs, or, as is usually the case, has been improvised with no planning at all.

Summary

A previous communication described an investigation in which the 3,555 Birmingham residents in mental hospitals in the period January 1 to March 31, 1957, were assigned to one of three classes according as they required for their care full, limited, or no hospital facilities. In the present report the data are used to assess the nature of the accommodation and staff which would be most appropriate for the patients in each class. It is suggested that the two considerations which have most bearing upon the arrangements are patients' mobility and the amount of supervision they require. These criteria have been used to divide the Birmingham mental hospital population into the nine groups tabulated below, which are drawn from a total population of approximately 1,100,000:

Hospital Facilities Required		Type of Accommodation	
		Traditional Ward	Hostel
Full	Closed Block	93	117
	Open Block	8	241*
Limited	Closed Block	298	1,066
	Open Block	59	1,232*
None	Open Block	—	441

* Although they required no supervision within the hospital, these patients were not considered suitable to leave the grounds unaccompanied.

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AN ANALYSIS OF FAMILIAL DATA ON PSYCHIATRIC PATIENTS: PARENTAL AGE, FAMILY SIZE, BIRTH ORDER, AND ORDINAL POSITION

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Introduction

Most mature students of human personality are agreed that normal and abnormal characteristics are determined by the complex interaction of numerous biological (*e.g.* genetic, neuro-endocrine, or metabolic), psychological, social, and cultural factors (Kluckhohn, Murray, and Schneider, 1953). However, there still exist wide differences of opinion as to the relative importance of various determinants in the causation of specific anomalies of personality and behaviour. While we are sometimes told that the nature-nurture controversy has long since been resolved, there are still many who view personality and its aberrations predominantly from either a genetical or an environmental aspect. The divergent opinions frequently represent at best no more than educated guesses, and at worst merely personal bias which results in unsolved problems being approached with preconceived convictions and consequent distortion of both observations and their interpretation.

Among the important consequences of differences in the orientation of the observer is the interpretation placed on studies of the families of mentally ill patients. There is much evidence of greatly increased incidence of mental illness among other members of the families of patients with certain mental disorders (*e.g.* Pollock, Malzberg, and Fuller, 1939; Kallmann, 1953). However, while some authorities maintain that such intra-familial concentration of these disorders represents mainly random genetic transmission (*e.g.* Kallmann, 1952, 1953; Slater, 1950, 1953), others regard transmission from parent to child as the result of harmful early experiences associated with intricate intra-familial relationships (*e.g.* Masserman, 1946; Lidz and Lidz, 1949; Weiss and English, 1949; Gerard and Siegel, 1950; Bowlby, 1952; Lidz, Parker and Cornelison, 1956).

In view of the observed concentration of mental disorders in the families of some patients, and the amount of speculation concerning both genetic transmission (Penrose, 1950) and functional interpersonal relationships or dynamics (Cleckley and Thigpen, 1955), it is at first sight remarkable how little is yet known about the structure or composition of the families of the mentally ill. It might perhaps be argued that without such information our investigations of function bear some similarity to the study of physiology in the absence of any knowledge of anatomy. However, the accumulation and analysis of data concerning the families of the mentally ill present serious technical difficulties and have often yielded barren results. Nevertheless, this is a most important area deserving the attention of both the biologist and the social scientist.

Among the familial studies that have been undertaken hitherto have been a number of investigations of parental age, family size, birth order, and ordinal position. Scientific interest in these variables developed during the latter part of the 19th century, in association with publications by Galton and other writers on heredity and eugenics. In the early years of the 20th century several quite extensive studies of family size and birth order were made, but early statistical techniques of analysis led to erroneous conclusions.

In their classic work on the determination of family size and of the distribution of characters in order of birth, Greenwood and Yule (1914) subjected to critical analysis earlier studies by Karl Pearson (1907), David Heron (1907), and Charles Goring (1913), which had appeared to indicate unduly large families, combined with an increased incidence in firstborn, in cases of tuberculosis, insanity, and criminality.

These latter findings had previously been criticized on various grounds by other writers' (Weinberg, 1910, 1913; Ploetz, 1913; Macaulay, 1912), and in their analysis of the data Greenwood

and Yule (1914) showed that the sizes of the families involved were not in fact unduly large, but if anything below average. They also failed to find any pathological weighting of the earlier born members of the sibships containing Heron's insane patients, although some degree of such weighting was evident among the sibships of tuberculous patients and criminals. In a series of imbeciles, which had been reported on by Pearson and other writers, they found a definite excess of first and second born children (amounting to about 10 per cent.), but also a proportionately greater excess of birth orders from the eighth onwards (amounting to over 40 per cent.). However, it is not clear to the present author whether this sample of imbeciles was drawn only from completed families, failing which the method of analysis would of itself tend to produce undue weighting in the later birth orders (McKeown and Record, 1956).

Pearson (1914) wrote at some length on the handicapping of the first born, and the impression has since remained that the latter are indeed more vulnerable with respect to the physical and mental abnormalities which are evident at birth or very early in life. An exception is provided by mongolism which occurs in the last born in about 50 per cent. of cases. However, this anomaly is also associated with advanced maternal age at birth of the affected child, and Penrose (1934) was able to demonstrate that the incidence of mongolism depends upon maternal age and not upon birth order (a conclusion reflected in a more recent statistical study by Malzberg, 1950). Achondroplasia occurs frequently in last born children, but is associated with increased age of the father rather than the mother. Anencephaly, on the other hand, shows no appreciable association with the age of either parent (Record and McKeown, 1949) and is more common in the early birth ranks. The interpretation of such findings has been discussed recently by Penrose (1955) and McKeown and Record (1956).

The studies on parental age and birth order outlined above have been of interest mainly to biologically-oriented scientists. Ordinal position in the family, on the other hand, has been of greater concern to those with a predominantly psychosocial conceptual framework. For several decades, theories relating ordinal position to personality characteristics, and to the incidence of neurosis and other disorders, have been associated in many persons' minds with the name of Adler. Unfortunately, Adler's rather dogmatic assertions were based on impressions derived from clinical experience rather than on scientific observation, and his views appear to have undergone some modification with the passage of time. Thus, in 1918, he stated that "the restless neurotics are, to a preponderating degree, second born children", while subsequently (quoted by Ansbacher and Ansbacher, 1956) he remarked that "in my experience the greatest proportion of problem children are oldest children; and close behind them come the youngest children".

Numerous other authors have added their own clinical impressions regarding the supposed vulnerability of some particular ordinal position. Thus Brill (1922) remarked that it would be best for the individual as well as the race that there should be no only children, while Hill (1945) asserted that a large proportion of eldest children develop into problem children, and also that a large number of youngest children fill the ranks of problem children, neurotics, and criminals.

Concurrently, there have been numerous attempts to relate ordinal position to various traits of personality, but when Murphy, Murphy, and Newcomb (1937) reviewed several hundred of these studies they found conflicting results. More recently attempts have been made to study ordinal position in relationship to the sex of the child, social class, and childhood experiences based on intra-familial interaction (*e.g.* Krout, 1939; Sears, 1950, 1957; Roberts and Myers, 1954).

The only articles with which the present author is familiar that include numerical comparisons of psychiatric disorders according to ordinal position (*e.g.* Norton, 1952; Wahl, 1954, 1956) use percentage figures, which depend in part upon the frequency distribution of family sizes in the sample. Estimates of mean family size reported in these articles also appear to be biased in the manner described by Greenwood and Yule (1914). However, Norton's study also includes interesting data on maternal age, birth order, and loss of a parent, and the data are so presented that further analysis is possible. In addition, Norton (1957) has been kind enough to provide supplementary unpublished data concerning both his series of psychiatric patients and of physically ill controls.

In the present article, it is planned to analyse further the data recorded by Norton (1952, 1957) on a large series of psychiatric patients (predominantly neurotic), and also certain data recorded by Malzberg (1938) concerning patients with dementia praecox and with manic-depressive psychoses, which he examined with regard to birth order (with negative findings).

Method

Sources of Data

Malzberg's cases consisted of 549 first admissions with dementia praecox, and 498 first admissions with manic-depressive psychoses, received at Manhattan State Hospital, New York, from 1930 to 1935. The patients were selected from the hospital records in the order of admission, but were excluded from tabulation when information was not sufficiently detailed, or when the patient was illegitimate, or had a half-sibling. The mean age at admission of patients with dementia praecox and with manic-depressive psychoses were 31·5 and 32·3 years respectively. These cases included both whites and Negroes, and Malzberg (1957) estimates the proportion of Negroes as somewhat in excess of 20 per cent.

Norton's 2,500 psychiatric patients were derived in two ways. 500 were a random sample of patients aged 15 years or more attending the psychiatric department of a general hospital (The London Hospital, England) during the period July, 1947, to July, 1948. Particulars of a further 2,000 patients who had attended the department between 1943 and 1947 were abstracted from their records. Most of the patients were suffering from psychoneuroses or personality disorders; those having florid psychoses and organic mental disorder were excluded, but the series contains a few mild endogenous depressives accounting for less than 10 per cent. of the total. The median age of the sample of 500 patients, calculated from unpublished data (Norton, 1957), was 30·4 years.

Norton's 500 controls were physically ill patients selected from the general in-patients of the hospital, who were matched for age and sex (and, less reliably, for social class). Patients referred for psychiatric consultation were rejected, as were those admitted for skin and neurological disorders. The period covered in collecting information about the control series was December, 1947, to March, 1949.

Since the publication of each of the two articles under consideration, detailed data has become available on the distribution of family sizes in the general populations from which the subjects were drawn (U.S. Bureau of the Census, 1943, 1947; Royal Commission on Population, 1954), and these sources of information have been used for comparative purposes in the present study.

Nature of Data

In the case of Malzberg's two series of psychotic patients, the data analysed here consist of Tables giving the numerical distribution of order of birth by size of family.

Similar Tables were utilized for both Norton's psychiatric patients and his controls, additional data being derived in this instance from Tables giving order of birth by maternal age and by paternal age, and also a Table comparing age differences between parents.

Comparisons Undertaken

Several types of comparison which were undertaken are listed below:

(*i*) Between maternal age, paternal age, and parental age differences, for 500 of Norton's psychiatric patients and his physically ill controls;

(*ii*) Between maternal ages for these two groups and for the general population of England and Wales;

(*iii*) Between observed and expected frequencies of birth order and ordinal position. This was based on a reconstruction of sibships by the Greenwood-Yule method (to examine family size and birth order) and an extension of this method (to examine ordinal position) for Norton's psychiatric patients and controls, and for both of Malzberg's series of patients;

(*iv*) Between family sizes of Norton's patients and controls, and of Malzberg's two groups of patients;

(*v*) Between family sizes for these various groups and the general populations from which they were drawn.

Greenwood-Yule Reconstruction of Sibships, and an Extension of this Method to examine Ordinal Position. —The method developed by Greenwood and Yule (1914) enables the calculation of corrected family sizes, and of expected frequencies of different birth orders, from samples taken through members of the sibships. The arithmetic involved is simple, but the method is not too widely known or understood, and it has been at times applied incorrectly as shown by McKeown and Record (1956). Some attempt at explanation might be considered desirable on these grounds alone, but in addition an extension of the method has here been used to examine ordinal position, which may require elucidation.

Table I shows the distribution of a model sample of affected subjects by size of sibship and birth order, and Table II shows the reconstruction of

the sibships of which the affected subjects are members. Let us consider separately certain aspects of the calculations concerning family size, birth order, and ordinal position.

The necessity of making a correction for **family size** depends on the fact that the likelihoood of meeting a member of a sibship of any given size in the general population is proportionate to the size of the sibship. Thus, if there were equal numbers of sibships containing x members and of sibships containing only one member, the chances of meeting individuals from sibships containing x members would be x times the chances of meeting only children (provided the sample is small in comparison with the total population). In order to obtain the approximately unbiased frequencies of family size that would have been obtained by taking a sample of *mothers*, it is therefore necessary to divide the biased frequencies obtained through members of the sibships (F_x), by the number of individuals in each sibship (x). Now the *biased* mean size of family is given by the formula $\Sigma(x\ F_x)/\Sigma(F_x)$. The unbiased or *corrected mean family* size is obtained by substituting (F_x/x) for (F_x) in the preceding formula, and is hence given by the formula $\Sigma(F_x)/\Sigma(F_x/x)$.

It is the distribution (F_x/x) for which other measures of central tendency, and standard deviation, should be derived. In calculating the *standard errors* of the mean, median, and standard deviation, however, there is some doubt whether the number of observations on which the standard error is based should be taken as $\Sigma(F_x)$ or as $\Sigma(F_x/x)$. Record (1957) considers that this number lies somewhere between the two, but that it is wiser to err on the conservative side by using the figure $\Sigma(F_x/x)$, so that estimates of the standard errors will be maximal (McKeown and Record, 1957), and this advice has been followed in calculating the standard errors given in the present study.

It is also the distribution (F_x/x) that must be examined when any comparison is made between sizes of family from which affected individuals came, and sizes of families in *samples obtained through mothers* (as in census figures on fertility). It is necessary to point out, however, that, in order to be comparable, other frequency distributions by size of family must refer to:

(*i*) *The same time period, locality, and socio-cultural group;*

(*ii*) *All children ever born* (not merely those still living at home, as recorded in some census statistics on housing and families);

(*iii*) *Completed families* (usually considered as the families of women over the age of 45);

(*iv*) *Children of mothers only* (figures for number of children ever born per woman reporting, or per married woman, are not comparable).

In the case of **birth order,** Greenwood and Yule calculated expected frequencies on the assumption that within each family of a given size each member or birth order should be affected equally frequently. The expected frequency of each order of birth then is obtained by summing the distribution (F_x/x) from the bottom upwards, The logical basis for this procedure should be apparent from examination of Tables I and II (overleaf). The expected frequency of each birth order may then be compared with the observed frequency by means of the χ^2 test.

To proceed now to the examination of **ordinal position** is very simple, since the distribution (F_x/x) gives the expected frequency of each specific ordinal position (*i.e.* eldest, youngest, and penultimate) for each size of family. Summing (F_x/x) from the bottom upwards (which has already been done in calculating expected frequencies of birth order) will therefore also give the expected total number in each specific ordinal position. Thus it appears that the expected frequency of eldest children (in all families having two or more children) is the same as that for youngest children, and both are the same as the expected frequency of children in the second birth order (E_2). The expected frequency of penultimate children (in all families having three or more children) is the same as that for children in the third birth order (E_3), and the expected frequency of intermediate children is obtained by differencing. In similar manner, the expected frequency of eldest, youngest, or penultimate children in families having x or more members is given by the frequency E_x (see Tables XII and XIII, pp. 54 and 55).

It will have been noted that *these calculations on ordinal position exclude the only child*, although the latter has hitherto been traditionally considered as an ordinal position (*e.g.* Glueck and Glueck, 1950; Norton, 1952; Wahl, 1954, 1956). However, it appears to the present author more appropriate to consider the frequency of only children in relation to the frequency distribution of family sizes (*vide supra*).

The present author has concluded that, when used with caution, the Greenwood-Yule method and the extension described are well suited to examining the sibships of psychiatric patients,

TABLE I

DISTRIBUTION OF SAMPLE OF AFFECTED SUBJECTS BY SIZE OF SIBSHIP AND BIRTH ORDER*
MODEL FOR DEMONSTRATING RECONSTRUCTION OF SIBSHIPS BY METHOD SHOWN IN TABLE II

Birth Order	Size of Sibship (x)						Totals = Observed Frequency of Each Birth Order (O_x)
	1	2	3	4	5	6	
1	a	b_1	c_1	d_1	e_1	f_1	$O_1\ (a+b_1+c_1+d_1+e_1+f_1)$
2		b_2	c_2	d_2	e_2	f_2	$O_2\ (b_2+c_2+d_2+e_2+f_2)$
3			c_3	d_3	e_3	f_3	$O_3\ (c_3+d_3+e_3+f_3)$
4				d_4	e_4	f_4	$O_4\ (d_4+e_4+f_4)$
5					e_5	f_5	$O_5\ (e_5+f_5)$
6						f_6	$O_6\ (f_6)$
Totals = Frequency of each Sibship of Size x ($=F_x$)	F_1 ($=a$)	F_2 ($=\Sigma b$)	F_3 ($=\Sigma c$)	F_4 ($=\Sigma d$)	F_5 ($=\Sigma e$)	F_6 ($=\Sigma f$)	$N\ (=\Sigma F_x=\Sigma O_x)$

*Dotted lines indicate ordinal positions (eldest, youngest, penultimate, and intermediate).

TABLE II

GREENWOOD-YULE RECONSTRUCTION OF FAMILY SIZE AND BIRTH ORDER OF MODEL SAMPLE OF AFFECTED SUBJECTS SHOWN IN TABLE I AND EXTENSION OF METHOD TO EXAMINE POSITION IN SIBSHIP

Size of Sibship (x)	Frequency of Sibship obtained from Sample of Affected Subjects (F_x)	Weighted Frequency (as would have been obtained from sample of mothers)* (F_x/x)	Expected Frequency of Each Birth Order (summing F_x/x from bottom upward) (E_x)	Observed Frequency of Each Birth Order (O_x)	$\frac{(O_x-E_x)^2}{E_x}$
1	$F_1\ (=a)$	$F_1/1\ (=a)$	$E_1\ (a+\bar{b}+\bar{c}+\bar{d}+\bar{e}+\bar{f})$	$O_1\ (a+b_1+c_1+d_1+e_1+f_1)$	..
2	$F_2\ (=\Sigma b)$	$F_2/2\ (=\bar{b})$	$E_2\ (\bar{b}+\bar{c}+\bar{d}+\bar{e}+\bar{f})$	$O_2\ (b_2+c_2+d_2+e_2+f_2)$	..
3	$F_3\ (=\Sigma c)$	$F_3/3\ (=\bar{c})$	$E_3\ (\bar{c}+\bar{d}+\bar{e}+\bar{f})$	$O_3\ (c_3+d_3+e_3+f_3)$	..
4	$F_4\ (=\Sigma d)$	$F_4/4\ (=\bar{d})$	$E_4\ (\bar{d}+\bar{e}+\bar{f})$	$O_4\ (d_4+e_4+f_4)$	..
5	$F_5\ (=\Sigma e)$	$F_5/5\ (=\bar{e})$	$E_5\ (\bar{e}+\bar{f})$	$O_5\ (e_5+f_5)$	..
6	$F_6\ (=\Sigma f)$	$F_6/6\ (=\bar{f})$	$E_6\ (\bar{f})$	$O_6\ (f_6)$	..
Totals ..	$\Sigma F_x\ (=N)$	$\Sigma(F_x/x)$	$\Sigma E_x\ (=\Sigma F_x=N)$	$\Sigma O_x\ (=\Sigma F_x=N)$	χ^2_x

Characteristics of Corrected Family Size (F_x/x)

Mode =
Median =
Mean =
Standard Deviation =

Goodness of Fit of Expected and Observed Frequencies of Birth Order

$\chi^2=\chi^2_p$
Degrees of Freedom $=n^1$
Probability $=p$

Ordinal Position in Sibship (of affected member)	Expected Frequency of Position (E_p)	Observed Frequency of Position (O_p)	$\frac{(O_p-E_p)^2}{E_p}$	Goodness of Fit for Position in Sibship
Eldest	$E_E\ (\bar{b}+\bar{c}+\bar{d}+\bar{e}+\bar{f})$	$O_E\ (b_1+c_1+d_1+e_1+\ _1)$	..	$\chi^2_p=$
Youngest	$E_y\ (\bar{b}+\bar{c}+\bar{d}+\bar{e}+\bar{f})$	$O_y\ (b_x+c_x+d_x+e_x+f_x)$	..	$n^1=3$
Penultimate	$E_p\ (\bar{c}+\bar{d}+\bar{e}+\bar{f})$	$O_{pu}\ (c_3+d_3+e_4+f_5)$	..	$p=$
Intermediate	$\Sigma E_x-(E_1+E_E+E_y)$	$\Sigma O_x-(a+O_E+O_y+O_{pu})$	..	
Totals	$\Sigma E_x-F_1\ (=N-a)$	$\Sigma O_x-F_1\ (=N-a)$	χ^2_p	

*F_x/x also constitutes the expected frequency distribution of each specific ordinal position in the sibship (*i.e.* eldest, youngest, or penultimate) by size of family. See also Table XII.

since the abnormality is not fully recognized until adult life, and hence:

(*i*) The sibships are complete, which is a prerequisite for application of the method (as otherwise the estimated family sizes are too small, and there is artificial weighting in late birth orders and ordinal positions);

(*ii*) Mental illness that is recognized in adult life does not apparently cause increased mortality during childhood.

(*iii*) Since the child that will later become mentally ill is not recognized as such by the parents, the latter cannot decide to limit the number of subsequent children on this account (as may occur in the case of some congenital abnormalities).

However, McKeown and Record (1957) have pointed out that the investigation of maternal age and birth order is extremely difficult in mental illness, because of the difficulty of assembling an acceptable control group, or of using the Greenwood Yule method in the case of conditions manifested over a wide age range. Moreover, there now appears to be some doubt whether birth rank comparisons are distributed as χ^2 (Halperin, 1953). A number of possible sources of error are discussed following the presentation of the results (*vide infra*).

THE RESULTS OF ANALYSIS ARE NOW PRESENTED AS FOLLOWS:

(*i*) The *parental ages* and age-difference of Norton's psychiatric patients are compared with those of his physically ill controls, and the median maternal ages of both groups are compared with figures of the general population of England and Wales;

(*ii*) The distribution of *family sizes* of Norton's psychiatric patients is compared with those of his controls and of the general population of England and Wales.
The distribution of *family sizes* of Malzberg's two groups of psychiatric patients are compared with each other and with selected samples of the population of New York City;

(*iii*) The expected and observed frequencies of *birth order* are compared for each of the four samples under analysis;

(*iv*) The expected and observed frequencies of different *ordinal positions* are examined for each of the four samples, for families of all sizes, and for families containing four or more members.

RESULTS

(1) PARENTAL AGE (Table III and Table IV, overleaf)

The *mean maternal age* of *Norton's psychiatric patients* exceeds that of his controls by 1·04 years, which is 2·58 times the standard error of the difference, so that the probability of this difference having occurred by chance is one in a hundred.

Comparison of the *median maternal ages* of both psychiatric patients and controls with the estimated median maternal age for women in England and Wales married in the period 1910–14 (given in the last column of Table IV as 29.79 years) shows good agreement for the controls, but again the difference for the maternal age of the psychiatric patients amounts to about 2·6 times its standard error.

In the case of *paternal age*, comparable figures were not available for the general population, and the nature of the data (Norton, 1957) did not enable the exact calculation of mean, standard deviation, and standard errors, but it may be noted that the median paternal age for patients exceeded that for controls by 0·96 years (as compared with 1·21 years difference between the recorded median ages of the mothers of these two groups).

The differences between the median ages of father and mother amounted to 2·08 years in the case of psychiatric patients, and 2·33 years in the case of controls. The latter figure corresponds closely with the figure of 2·3 years given by Penrose (1955) as the difference in mean parental ages for the population of England and Wales in 1939.

TABLE III

MATERNAL AGE AT BIRTH OF 500 OF NORTON'S PSYCHIATRIC PATIENTS (PREDOMINANTLY NEUROTIC) AND OF 500 CONTROLS (PHYSICALLY ILL PATIENTS MATCHED FOR AGE, SEX, AND SOCIAL CLASS), THE LONDON HOSPITAL, ENGLAND, 1947–49 (a) (b) (c)

Characteristics of Maternal Age (yrs)	Psychiatric Patients	Physically Ill Controls
Median	30·74 ± 0·36	29·53 ± 0·36
Mean	31·14 ± 0·28	30·10 ± 0·29
Standard Deviation ..	6·35 ± 0·20	6·42 ± 0·20

Difference between the Two Means: 1·04 ± 0·404; $t = 2{\cdot}58$; $p < 0{\cdot}01$

NOTES

(a) *Median age of father at birth* was 32·82 years for 500 of Norton's psychiatric patients, and 31·86 years for his 500 controls (calculated from unpublished data, 1957).

(b) *Difference between median ages of father and mother* was therefore 2·08 years for Norton's psychiatric patients and 2·33 years for his controls. The latter corresponds with 2·3 years for the differences in mean maternal ages for the general population of England and Wales, 1939 (Penrose, 1955).

(c) *Mother one or two years older than father* in 74 out of 500 of Norton's psychiatric patients as compared with 47 out of 500 controls. Difference = 5·4 per cent. ± 2·02; $t = 2{\cdot}67$; $p < 0.01$.

TABLE IV

AGES OF WOMEN AT FIRST MARRIAGE AND WHEN HALF OF ALL CHILDREN HAD BEEN BORN, BY DATE OF MARRIAGE, ENGLAND AND WALES, 1900–24.

Date of Marriage	Mean Ages at Marriage by Broad Social Status Groups(a)			Median Ages at Marriage(b)	Median Duration of Marriage when Half of All Children had been Born(c)	Median Age of Mother when Half of All Children had been Born(d) (add two preceding columns)
	Non-Manual	Manual	Both Groups			
1900–09	25·39	24·17	24·61	24·10	5·75	29·85
1910–14	27·76	24·46	24·94	24·40	5·39	29·79(e)
1915–19	25·79	24·77	25·15	24·49	4·93	29·42
1920–24	25·57	24·33	24·78	24·21	4·50	28·71

(a) Royal Commission on Population, Papers Volume VI, The Trend and Pattern of Fertility in Great Britain (1954) pt. I, p. 105 (Table 33), H.M.S.O., London.
(b) *Ibid.*, pt. I, p. 84 (interpolated from Table 13).
(c) *Ibid.*, pt. I, p. 162 (interpolated from Table 1).
(d) Corresponds closely with mean maternal age of 28·6 years for England and Wales, 1939 (Penrose, 1954).
(e) Median age of Norton's psychiatric patients and of his physically ill controls was 30·4 years by 1947–49 (calculated from unpublished data, 1957). It is estimated that the median date of marriage for the mothers of both groups was approximately 1912. The median maternal age of the general population married around this time corresponds closely with that of Norton's physically ill controls (see Table III).

Norton's unpublished data (1957) also indicate that 74 out of 500 psychiatric patients, as compared with 47 out of 500 controls, had *mothers one or two years older than their fathers*. This difference amounts to 5·4 per cent. which is 2·67 times its standard error, so that the probability of its being due to chance is less than one in a hundred.

(2) FAMILY SIZE (Tables V, VI, VII, VIII, IX, X, and XI)

Characteristics of family size for *Norton's psychiatric patients* (Table V, opposite) show no significant differences from those of his physically ill controls (Table VI, overleaf).

The mean size of family for both these samples is significantly less than the mean size of family for all social status groups of mothers in England and Wales married in the period 1910–14, but is well within the limits of family size differences between the two broad status groups, being weighted on the side of the non-manual group (see Tables VII and VIII, overleaf).

However, the distributions of both Norton's psychiatric patients and his physically ill controls show an unusual feature—namely a mode of unity. Now it may be seen from Table VIII that a mode of one was not found in marriages of the appropriate duration, except in the case of marriages taking place when the mother was over the age of 30 Moreover, no family recorded in Table VIII that had a mode of only one had a mean exceeding 2·6 (whereas the means of the two samples under discussion were approximately 3·1).

It appears therefore that both Norton's samples contain an unduly high proportion of families in which the affected member was an only child.

Both Malzberg's series of patients, on the other hand (see Tables IX and X, pp. 52 and 53), show distributions by size of family (F_x/x) that agree reasonably well with what might be expected after consideration of their mixed white and Negro composition, and after comparison with available figures for New York City (see Table XI, p. 54).

However, there is a suggestive difference of 0·5 between the mean family sizes of Malzberg's two groups of patients, which amounts to 1·69 times its standard error, and which could therefore be attributed to chance with a probability of slightly less than one in ten.

(3) BIRTH ORDER (Tables V, VI, IX, and X)

This variable was investigated in the original studies by both Malzberg (1938) and Norton (1952), and nothing need to be added to their own analyses. It was, however, necessary to calculate expected frequencies of each birth order for the purposes of the present article (as a preliminary to anaylsis of ordinal position), and it was relatively easy to apply the χ^2 test to expected and observed frequencies of birth order.

In *Norton's psychiatric patients* (Table V, opposite), there was a lower observed incidence than expected in was a lower observed incidence than expected in the early birth ranks and a moderately increased incidence was observed in the later ranks, findings which could have occurred on the basis of chance less than once in fifty trials. These findings are

TABLE V

RECONSTRUCTION OF SIBSHIPS OF NORTON'S 2,500 PSYCHIATRIC PATIENTS (PREDOMINANTLY NEUROTIC), THE LONDON HOSPITAL, ENGLAND, 1943–48(a)

Size of Sibship (x)	Frequency of Sibship obtained from Sample of Affected Subjects (F_x)	Weighted Frequency (as would have been obtained from sample of mothers) (F_x/x)	Expected Frequency of Each Birth Order (summing F_x/x from bottom upward) (E_x)	Observed Frequency of Each Birth Order (O_x)	$\frac{(O_x-E_x)^2}{E_x}$
1	226	226·0	794·68	751	2·58
2	358	179·0	568·68	553·5	0·32
3	374	124·67	389·68	369	1·10
4	348	87·0	265·01	266·5	0·01
5	335	67·0	178·01	207	4·72
6	225	37·5	111·01	121	0·90
7	178	25·43	73·51	87	2·49
8	139	17·38	48·08	52	0·32
9	118	13·11	30·70	34	0·36
10	84	8·4	17·59	24	2·31
11	33	3·0	9·19	15	
12	30	2·5	6·19	5	
13	25	1·92	3·69	6	
14	9	0·65	1·77	2	
15	9	0·6	1·12	4	6·10
16	2	0·12	0·52	1	
17	3	0·18	0·40	2	
18	3	0·17	0·22	0	
19	1	0·05	0·05	0	
Totals	2,500	794·68	2,500·10	2,500	21·23

Characteristics of Corrected Family Size (F_x/x)

Mode = 1
Median = 1·96 ± 0·10
Mean = 3.14 ± 0·08
Standard Deviation = 2·34 ± 0·06

Goodness of Fit of Expected and Observed Frequencies of Birth Order

$\chi^2 = 21·23(a)$
$n^1 = 10$
$\therefore p < 0·02$

Ordinal Position in Sibship (of affected member)	Expected Frequency (E_p)	Observed Frequency (O_p)	$\frac{(O_p-E_p)^2}{E_p}$	Goodness of Fit for Position in Sibship
Eldest	568·68	525	3·38	
Youngest	568·68	639	8·7	$\chi^2 = 15·88(a)$
Penultimate	389·68	409	0·96	$n^1 = 3$
Intermediate	746·96	701	2·84	$\therefore p < 0·01$
Totals	2,274·00	2,274	15·88	

(a) This sample is approximately five times as large as each of the other three samples analysed here. Hence figures for χ^2 given in Table V (relating to distributions by birth order and ordinal position) are also five times as large as would have been obtained from equivalent distributions in a sample of 500 patients.

in contrast to those for his series of controls, which showed remarkably good fit between observed and expected values (Table VI, overleaf), but may be at least partly attributable to the much larger size of the sample of psychiatric patients examined here.

Expected and observed frequencies of birth order also showed no significant differences in *either of Malzberg's groups of patients* (Tables VIII and IX, pp. 51 and 52).

(4) ORDINAL POSITION (Tables V, VI, IX, X, XII, and XIII)

Here there are a number of significant findings.

Norton's psychiatric patients (Table V) exhibit a poor fit of observed and expected frequencies that could have resulted from chance only once in several hundred trials (there being a moderate excess of observed over expected frequency of youngest children, with a relative deficiency of eldest and intermediate). Norton's controls (Table VI, overleaf), on the other hand, show a remarkably good fit between the frequencies observed and those expected (although part of this difference may be attributable to the much larger sample of psychiatric patients examined here).

Turning to *Malzberg's patients with dementia praecox* (Table IX, p. 52), there again appears to be an unexpectedly high frequency of youngest children, but in this instance it is not statistically significant. However, in *Malzberg's manic-depressive patients* there is an extremely significant distribution (a relative excess of eldest and youngest associated with a deficiency of penultimate children).

In order to examine ordinal position in relation to family size, two further Tables were constructed.

TABLE VI

RECONSTRUCTION OF SIBSHIPS OF NORTON'S 500 PHYSICALLY ILL CONTROLS, THE LONDON HOSPITAL, ENGLAND, 1947–49(a)

Size of Sibship (x)	Frequency of Sibship obtained from Sample of Affected Subjects (F_x)	Weighted Frequency (as would have been obtained from sample of mothers) (F_x/x)	Expected Frequency of Each Birth Order (summing F_x/x from bottom upward) (E_x)	Observed Frequency of Each Birth Order (O_x)	$\frac{(O_x-E_x)^2}{E_x}$
1	42	42·0	160·98	159	0·025
2	78	39·0	118·98	121	0·033
3	95	31·67	79·98	89·5	1·13
4	74	18·5	48·31	42·5	0·72
5	39	7·8	29·81	27	0·27
6	47	7·83	22·01	19·5	0·28
7	36	5·14	14·18	14·5	0·01
8	23	2·87	9·04	9	
9	22	2·44	6·17	7	
10	11	1·1	3·73	2·5	
11	11	1·0	2·63	4·5	
12	6	0·5	1·63	1	
13	9	0·69	1·13	1	0·09
14	2	0·14	0·44	0	
15	2	0·13	0·30	1	
16	1	0·06	0·17	0	
17	1	0·06	0·11	1	
18	0	0·0	0·05	0	
19	1	0·05	0·05	0	
Totals	500	160·98	499·70	500	2·56

Characteristics of Corrected Family Size (F_x/x)

Mode = 1
Median = 1·99 ± 0·22
Mean = 3·10 ± 0·18
Standard Deviation = 2·30 ± 0·13

Goodness of Fit of Expected and Observed Frequencies of Birth Order

$\chi^2 = 2·56$
$n^1 = 7$
$\therefore p > 0·9$

Ordinal Position in Sibship (of affected member)	Expected Frequency (E_p)	Observed Frequency (O_p)	$\frac{(O_p-E_p)^2}{E_p}$	Goodness of Fit for Position in Sibship
Eldest	118·98	117	0·03	
Youngest	118·98	123·5	0·17	$\chi^2 = 0·24$
Penultimate	79·98	78·5	0·03	$n^1 = 3$
Intermediate	140·06	139	0·01	$\therefore p > 0·95$
Totals	458·0	458·0	0·24	

(a) Calculated from Norton's unpublished data (1957).

TABLE VII

NUMBER OF LIVE CHILDREN EVER BORN (BY 1946) PER MARRIED WOMAN REPORTING, AND PER MARRIED MOTHER, BY DATE OF MARRIAGE, ENGLAND AND WALES(a)

Date of Marriage	Mean Number of Live Births per Married Woman (childless or mother)	Percentage of Married Women remaining Childless	Number of Live Births per Married Mother		
			Mean	Median	Mode
Before 1890	5·14	6·8	5·51	4·52	3
1890–99	4·34	8·1	4·73	3·61	2=3
1900–09	3·53	10·2	3·93	2·80	2
1910–14	2·98	12·3	3·40	2·24	2
1915–19	2·60	14·6	3·05	1·97	2
1920–24	2·41	14·8	2·83	1·79	2

(a) Royal Commission on Population, Papers Volume VI, The Trend and Pattern of Fertility in Great Britain, 1954, pt. I, p. 81 (extracted and calculated from Table 7). H.M.S.O., London.

In Table XII, (p. 54), expected frequencies of specific ordinal positions for each size of sibship may be compared directly with observed values. From examination of this Table it appeared that there might be greater differences between incidence according to ordinal position in large families than in small. Hence comparisons were made between total expected and observed frequencies of ordinal position among *patients from sibships having four or more members*. The results (Table XIII, p. 55) are of great interest. They reveal a statistically significant lack of correspondence between observed and expected frequencies in all three types of psychiatric patient, although there is once more a good fit in the case of Norton's controls. In all three groups of psychiatric patients, however, the observed frequency of youngest children is unduly

TABLE VIII

MEAN AND MODAL NUMBER[a] OF LIVE CHILDREN EVER BORN (BY 1946) PER MARRIED MOTHER, BY YEAR OF MARRIAGE, AGE AT MARRIAGE, AND BROAD SOCIAL STATUS GROUP, ENGLAND AND WALES, 1900–24[b]

Year of Marriage ..		1900–09		1910–14		1915–19		1920–24	
Social Status Group ..		Manual	Non-Manual	Manual	Non-Manual	Manual	Non-Manual	Manual	Non-Manual
Age at Marriage (yrs)	Under 20	5·7 (4)	4·4 (3)	5·0 (4)	3·8 (3)	4·4 (2)	3·3 (2)	3·9 (2)	3·1 (2)
	22½–25	4·0 (2)	3·2 (2)	3·6 (2)	2·8 (2)	3·3 (2)	2·6 (2)	2·8 (2)	2·3 (2)
	25 –27½	3·5 (2)	2·9 (2)	3·1 (2)	2·5 (2)	2·8 (2)	2·3 (2)	2·6 (1)	2·1 (2)
	27½–30	3·2 (2)	2·7 (2)	2·9 (2)	2·4 (2)	2·7 (2)	2·1 (1)	2·4 (1)	2·0 (1)
	30 –32½	2·9 (2)	2·6 (1)	2·6 (1)	2·3 (1)	2·5 (1)	2·1 (1)	2·3 (1)	2·0 (1)
	40 –45[c, d]	2·5 (1)	1·7 (1)	2·0 (1)	1·6 (1)	2·0 (1)	1·6 (1)	1·9 (1)	1·6 (1)
	All Ages under 45	4·36 (2)	3·21 (2)	3·76 (2)	2·77 (2)	3·36 (2)	2·50 (2)	3·04 (2)	2·29 (2)
All Ages under 45 and All Social Status Groups		3·93 (2)		3·40 (2)		3·05 (2)		2·83 (2)	

(a) Mode given in brackets (in italics). Dotted line separates families of modal size 1 from those with a mode of 2 or more. Note that with a mode of 1 the mean does not exceed 2.6

(b) Royal Commission on Population, Papers Volume VI. The Trend and Pattern of Fertility in Great Britain, 1954, pt. II, p. 66–73, 120–129, and 138–147 (calculated from Tables C. 10, 20, 30, and 40, and from among Tables G. 1–40 and H. 1–40). H.M.S.O. London,

(c) Percentage of married women remaining childless rises with increasing age at marriage. Ratio of mean number of live births per mother (tabulated above) to mean number of live births per woman reporting (childless as well as mothers) depends on the percentage of women remaining childless, and hence also rises with increasing age at marriage.

(d) Modal number of live births for all married women reporting (childless as well as mothers) was zero for all ages at marriage from 32½ yrs. upward (for both broad social status groups, and throughout the period 1900–24).

high, and in both Malzberg's series it was approximately double the observed frequency recorded for penultimate children (although the expected frequencies in this instance are identical).

DISCUSSION

(1) POSSIBLE SOURCES OF ERROR IN RESULTS

There are several possible types of bias which require consideration before any tentative interpretations are placed upon the results that have been presented.

First, the question might be raised whether the physically ill patients selected by Norton provide suitable controls for his series of psychiatric patients —particularly in view of recent findings indicating a high incidence of psychiatric abnormality in a random sample of surgical ward patients (Zwerling, Titcher, Gottschalk, Levine, Culbertson, Cohen, and Silver, 1955; Titchener, Zwerling, Gottschalk, Levine, Culbertson, Cohen, and Silver, 1956). However, it may have been noted that Norton's physically ill controls were *selected* by age, and to a considerable extent also by diagnosis (both on account of the age selection, and by exclusion of neurological dermatological and any suspected psychiatric disorders), from among the general in-patients of the hospital. Moreover, they closely approach the statistical expectations for characteristics under examination in the present study. Thus, their median maternal age and their mean family size correspond closely with those estimated for mothers in the general population marrying in the period 1910–1914; the difference between the median ages of their fathers and mothers agrees with the difference between the mean parental ages for the control population given by Penrose (1955); their observed frequency distributions by birth order and by ordinal position closely resemble the frequencies expected on the basis of chance. Their only anomalous features, indeed, are an unexpectedly high proportion of only children, and 20 per cent. excess of observed over expected frequency of youngest children in families having four or more members (both of which observations were found also in the psychiatric patient group).

The other possible sources of bias considered here concern the Greenwood-Yule method of examining family size and birth order, and the extension which has been introduced in order to examine ordinal position.

It has been mentioned earlier that this method is inapplicable unless the sibships containing the affected member have been completed, as otherwise the estimate of mean family size is too low, and there is an apparent excess in the larger (later)

TABLE IX

RECONSTRUCTION OF SIBSHIPS OF MALZBERG'S 549 PATIENTS WITH DEMENTIA PRAECOX, MANHATTAN STATE HOSPITAL, NEW YORK, 1930–35

Size of Sibship (x)	Frequency of Sibship obtained from Sample of Affected Subjects (F_x)	Weighted Frequency (as would have been obtained from sample of mothers) (F_x/x)	Expected Frequency of Each Birth Order (summing F_x/x from bottom upward) (E_x)	Observed Frequency of Each Birth Order (O_x)	$\frac{(O_x-E_x)^2}{E_x}$
1	27	27·0	154·66	153	0·02
2	73	36·5	127·66	126	0·02
3	95	31·67	91·16	96	0·26
4	69	17·25	59·49	59	0·00
5	67	13·4	42·24	34	1·61
6	65	10·83	28·84	24	0·81
7	43	6·15	18·01	25	2·72
8	45	5·62	11·86	19	4·30
9	21	2·33	6·24	4	
10	20	2·0	3·91	3	
11	9	0·82	1·91	2	
12	3	0·25	1·09	2	
13	5	0·38	0·84	0	
14	4	0·29	0·46	1	
15	0	0·0	0·17	1	0·29
16	1	0·06	0·17	0	
17	1	0·06	0·11	0	
18	0	0·0	0·05	0	
19	0	0·0	0·05	0	
20	0	0·0	0·05	0	
21	1	0·05	0·05	0	
Totals	549	154·66	549·02	549	10·03

Characteristics of Corrected Family Size (F_x/x)

Mode = 2
Median = 2·49 ± 0·24
Mean = 3·55 ± 0·19
Standard Deviation = 2·36 ± 0·13

Goodness of Fit of Expected and Observed Frequencies of Birth Order

$\chi^2 = 10·03$
$n^1 = 8$
$\therefore p < 0·30$

Ordinal Position in Sibship (of Affected member)	Expected Frequency (E_p)	Observed Frequency (O_p)	$\frac{(O_p-E_p)^2}{E_p}$	Goodness of Fit for Position in Sibship
Eldest	127·66	126	0·02	$\chi^2 = 4·91$
Youngest	127·66	145	2·35	$n^1 = 3$
Penultimate	91·16	76	2·52	$\therefore p < 0·20$
Intermediate	175·52	175	0·00	
Totals	522	522	4·91	

birth orders (particularly the last born of small families). In all the four series of cases examined in the present study, the mean age was in the neighbourhood of 30 years, and the youngest known member was 15 years of age, so that these sibships may be regarded as very probably complete. However, there might be a slight under-representation of the earlier born members of large families (these being recorded as slightly smaller than their ultimate size), and hence a small relative excess of the later born members of large families (affecting the higher birth orders and penultimate as well as youngest children).

Malzberg (1938) felt that there was a great probability of the observed number of first born being in excess of the true total, because a number of the patients might have lost older siblings in infancy and grown up in ignorance of such events. It might be argued that such individuals grew up psychologically as first born and should rightly be considered as such, but in the present instance the matter is largely academic since it is an excess of younger rather than older members of sibships that has to be accounted for.

A further possible source of bias suggested by Malzberg (1938) concerned the assumption that, since the (cumulative) rate of mental illness advances with age, this would lead to an apparent excess of earlier born among the patients. The present author does not believe that this problem arises, provided that the recording of a given sibship depends upon the affected member being diagnosed or admitted to hospital within a given time period, and also provided that the recording of the affected member does not in any way lead to the recording of any other member of the sibship affected before

TABLE X

RECONSTRUCTION OF SIBSHIPS OF MALZBERG'S 498 PATIENTS WITH MANIC-DEPRESSIVE PSYCHOSES, MANHATTAN STATE HOSPITAL, NEW YORK, 1930–35

Size of Sibship (x)	Frequency of Sibship obtained from Sample of Affected Subjects (F_x)	Weighted Frequency (as would have been obtained from sample of mothers) (F_x/x)	Expected Frequency of Each Birth Order (summing F_x/x from bottom upward) (E_x)	Observed Frequency of each Birth Order (O_x)	$\frac{(O_x-E_x)^2}{E_x}$
1	16	16·0	122·89	140	2·40
2	45	22·5	106·89	101	0·33
3	63	21·0	84·39	78	0·49
4	73	18·25	63·39	62	0·03
5	79	15·8	45·14	37	1·47
6	71	11·83	29·34	37	2·00
7	48	6·86	17·51	15	0·36
8	36	4·5	10·65	9	0·26
9	21	2·33	6·15	7	
10	10	1·0	3·82	1	
11	9	0·82	2·82	2	
12	10	0·83	2·00	1	
13	7	0·54	1·17	2	
14	5	0·36	0·63	3	
15	1	0·07	0·27	0	
16	1	0·06	0·20	0	0·01
17	0	0·0	0·14	1	
18	0	0·0	0·14	0	
19	1	0·05	0·14	1	
20	0	0·0	0·09	0	
21	0	0·0	0·09	1	
22	1	0·05	0·09	0	
23	1	0·04	0·04	0	
Totals	498	122·89	497·99	498	7·35

Characteristics of Corrected Family Size (F_x/x)

Mode = 2
Median = 3·11 ± 0·28
Mean = 4·05 ± 0·23
Standard Deviation = 2·52 ± 0·16

Goodness of Fit Expected and Observed Frequencies of Birth Order

$\chi^2 = 7·35$
$n^1 = 8$
$\therefore p < 0·50$

Ordinal Position in Sibship (of Affected Member)	Expected Frequency (E_p)	Observed Frequency (O_p)	$\frac{(O_p-E_p)^2}{E_p}$	Goodness of Fit for Position in Sibship
Eldest	106·89	124	2·75	$\chi^2 = 16·58$
Youngest	106·89	124	2·75	$n^1 = 3$
Penultimate	84·39	54	11·0	$\therefore p < 0·001$
Intermediate	183·83	180	0·08	
Totals	482	482	16·58	

that time. Again, this possible form of bias would result in apparent weighting of earlier and not later born members of sibships.

Norton (1957) raises a valid difficulty when he points out that apparent weighting of younger members of large families may result from diminishing numbers of large families in the general population (it therefore being relatively somewhat more likely that we should meet a younger member of a large family started, let us say, before the end of the 19th century, than that we should meet an older member of a family of the same size started at a later date). In addition to causing an apparent weighting of youngest members of large families, this would of course lead to a relative weighting of penultimate members, and also a relative weighting of higher birth orders as a whole—neither of which was found in the case of Norton's physically ill controls (see Tables VI and XIII) or in either of Malzberg's series (Tables IX, X, and XIII).

The other possible source of error that will be examined here concerns the number of cases encountered during the recording of data for which birth order and/or size of family could not be ascertained. It appears possible that such "unascertained" cases might be more likely to belong to large families and intermediate positions. Some light may be thrown on this problem by examining the distribution of "unascertained" cases recorded by Malzberg (1950) in a statistical analysis of mongols in the registers of the New York State schools for mental defectives on April 1, 1948. Out of 880 mongols so recorded, the birth order alone could not be ascertained in 95 cases, the size of family alone in only four cases, and neither birth order nor family size in a further 23 cases. The total number

TABLE XI

PERCENTAGE DISTRIBUTION BY SIZE OF COMPLETED FAMILY, FOR MOTHERS OF MALZBERG'S PSYCHOTIC PATIENTS (ADMITTED TO MANHATTAN, STATE HOSPITAL, NEW YORK, 1930-35), AND FOR MOTHERS AGED 50 TO 74 YEARS LIVING IN NEW YORK CITY, 1940(a)

Characteristics of Sample	Mean Number of Children ever Born per Mother	Percentage of Mothers with Number of Children (completed families)									
		1	2	3	4	5	6	7	8	9	10 and Over
Mothers(b) of Malzberg's Patients with Dementia Praecox (F_x/x)	3·55(c)	17·5	23·6	20·7	11·2	8·7	7·1	4·0	3·6	1·5	2·5
Mothers(b) of Malzberg's Manic-Depressives(F_x/x)	4·05(c)	13·0	18·3	17·1	14·8	12·8	9·6	5·6	3·6	1·9	3·1
White Mothers, New York City, aged 50–74 in 1940(d)	3·60	18·0	22·1	18·6	14·6	9·4	6·0	3·9	2·7	1·9	3·0
Non-White Mothers, New York City, aged 50–74 in 1940(d)	3·35	24·0	25·8	17·3	7·5	7·0	6·2	2·8	4·4	2·1	2·8

(a) U.S. Bureau of the Census (1943), 16th Census of the United States 1940, Population – Differential Fertility 1940 and 1910, Fertility for States and Large Cities, Washington, D.C., U.S. Government Printing Office, Pp. 222 and 226 (calculated from Tables 40 and 41).

(b) Mean ages of Malzberg's patients on admission (1930–35) were 31·5 years (dementia praecox) and 32·3 years (manic-depressives). If it is assumed that the mean ages of their mothers at marriage was 25, and at the time of the patient's birth 30 years, then the mean date of their mother's marriage would have been 1895, and the mean ages of their mothers in 1940 (if still living) would have been 70 years.

(c) The difference between the mean number of children in the sibships of Malzberg's patients with dementia praecox and of the manic-depressives is $0·50 \pm 0·296$. Hence $t = 1·69$; $p < 0·10$.

(d) Figures for the fertility of women in the United States by *date* of marriage were not published, nor were figures for the fertility of women in New York City by *duration* of marriage in 1940. However, the figures tabulated above for white mothers, New York City, aged 50–74 in 1940, are very similar to others calculated for white mothers, North-eastern States, aged 65–69 and married for 40–49 years in 1940, and also for those aged 70–74 and married for 45–54 years in 1940 (U.S. Bureau of the Census, 1947).

TABLE XII

DISTRIBUTION OF EXPECTED AND OBSERVED FREQUENCIES OF PATIENTS BY ORDINAL POSITION AND SIZE OF SIBSHIP.

NORTON'S 2,500 PSYCHIATRIC PATIENTS AND 500 PHYSICALLY ILL CONTROLS. MALZBERG'S 549 PATIENTS WITH DEMENTIA PREACOX AND 498 MANIC-DEPRESSIVES(a)

Ordinal Position			Size of Sibship										Totals(b)
			2	3	4	5	6	7	8	9	10	Over 10	
Norton's Psychiatric Patients	Expected (F_x/x) ..		*179*	*124·7*	*87*	*67*	*37·5*	*25·4*	*17·4*	*13·1*	*8·4*	*9·2*	*568·68*
	Observed	Eldest	172	124	89	41	36	24	11	11	6	11	525
		Youngest	186	122	91	89	41	35	21	18	17	19	639
		Penultimate	—	128	77·5	73·5	40	32	22	14	7	15	409
Norton's Physically Ill Controls	Expected (F_x/x)		*39*	*31·7*	*18·5*	*7·8*	*7·8*	*5·1*	*2·9*	*2·4*	*1·1*	*2·6*	*118·98*
	Observed	Eldest	49	23	11	6	12	6	2	4	1	3	117
		Youngest	29	36	20·5	10	5·5	7	4	5	1	5·5	123·5
		Penultimate	—	36	21·5	6	3·5	5	1	3	0	2·5	78·5
Malzberg's Patients with Dementia Praecox	Expected (F_x/x)		*36·5*	*31·7*	*17·2*	*13·4*	*10·8*	*6·1*	*5·6*	*2·3*	*2·0*	*1·9*	*127·66*
	Observed	Eldest	41	30	16	15	10	4	6	0	3	1	126
		Youngest	32	32	24	17	13	9	12	1	2	3	145
		Penultimate	—	33	10	7	8	4	4	4	2	4	76
Malzberg's Manic-Depressives	Expected (F_x/x)		*22·5*	*21*	*18·2*	*15·8*	*11·8*	*6·9*	*4·5*	*2·3*	*1·0*	*2·8*	*106·89*
	Observed	Eldest	27	25	17	24	14	7	7	2	0	1	124
		Youngest	18	23	16	15	23	8	6	5	1	9	124
		Penultimate	—	15	17	11	4	3	1	2	0	1	54

(a) See also Tables V, VI, IX, X, and XIII

(b) Totals for sibships of two or more children (three or more children, in the case of penultimate patients)

TABLE XIII

EXPECTED AND OBSERVED FREQUENCIES OF ORDINAL POSITIONS AMONG PATIENTS *FROM SIBSHIPS HAVING FOUR OR MORE MEMBERS*

NORTON'S PSYCHIATRIC PATIENTS AND PHYSICALLY ILL CONTROLS
MALZBERG'S PATIENTS WITH DEMENTIA PRAECOX AND MANIC-DEPRESSIVE PSYCHOSES(*a*)

Ordinal Position in Sibship	Expected Frequency in Model Sample(*b*)	Norton's Psychiatric Patients(*c*)		Norton's Physically Ill Controls		Malzberg's Patients with Dementia Praecox		Malzberg's Manic-Depressives	
		Expected	Observed	Expected	Observed	Expected	Observed	Expected	Observed
Eldest	E_1	265·01	229	48·31	45	59·49	55	63·39	72
Youngest ..	E_4	265·01	331	48·31	58·5	59·49	81	63·39	83
Penultimate ..	E_3	265·01	281	48·31	42·5	59·49	43	63·39	39
Intermediate ..	$\Sigma E_X-(E_1+E_3+E_4)$	746·97	701	140·07	139	175·53	175	183·83	180
Totals ..	$\Sigma E_X-(F_1+F_3+F_4)$	1,542	1,542	285	285	354	354	374	374
Goodness of Fit of Expected and Observed Ordinal Positions		$\chi^2=25·2$(*c*) $n^1=3$ $\therefore p<0·001$		$\chi^2=3·06$ $n^1=3$ $\therefore p<0·50$		$\chi^2=12·7$ $n^1=3$ $\therefore p<0·01$		$\chi^2=16·7$ $n^1=3$ $\therefore p<0·001$	

(*a*) See also Tables V, VI, IX, X, and XII
(*b*) See Tables I and II
(*c*) The sample of Norton's psychiatric patients examined here is approximately five times as large as each of the other three samples. Hence figures for χ^2 given in Table V, and above, are also five times as large as would have been obtained from equivalent distributions in a sample of 500 patients.

of mongols in which birth order remained unascertained was therefore 118, and their distribution by size of family is shown in Table XIV. This Table also shows the distribution by family size of the estimated total numbers of mongols in whom birth order was unascertained (after redistribution of cases in which family size was unascertained), and the expression of the latter as a percentage of all the mongols in each family size.

This Table shows that the proportion of cases in which birth order was not ascertained tended to increase with the size of family, and it was also found that, for families having four or more members, the ratio of cases in which birth order was unascertained to those in whom it was established was about 1:4. If it were assumed that all cases in which birth order was unascertained were either intermediate or penultimate, for families having four or more members the ratio of unascertained penultimate to unascertained intermediate (if evenly distributed) would have been approximately 1 : 3. If these figures were applied to Malzberg's patients with dementia praecox and manic-depressive psychoses from families with four or more members (Table XIII), the result would be about ninety additional unascertained cases in either group, and of these about 24 could have been penultimate. Hence, even on the assumption that all unascertained cases would be intermediate or penultimate children, the marked difference between observed incidence in youngest and penultimate in these two groups would not be nearly eliminated. In fact, however, this assumption is clearly unjustified, since, even in families having only two members (*i.e.* only eldest and youngest), the birth order was unascertained in at least ten out of 186 cases. This source of error, therefore, is one which requires consideration in future studies of this nature, but one which could not have been responsible for certain differences in incidence according to ordinal position noted in the present analysis.

TABLE XIV

DISTRIBUTION BY SIZE OF FAMILY OF CASES IN WHICH BIRTH ORDER WAS NOT ASCERTAINED, IN A SERIES OF MONGOLS ANALYSED BY MALZBERG (1950)

Size of Family	1	2	3	4	5	6	7	8	9	10 and Over	Not Known	Totals
Total Number of Mongols	72	186	174	109	89	83	53	24	17	46	27	880
Number of Mongols for whom Birth Order was Unascertained	0	10	13	16	10	15	12	8	2	9	23	118
Estimated Number of Mongols for whom Birth Order was Unascertained, if all Family Sizes were Known(*a*)	0	12·4	16·1	19·8	12·4	18·6	14·9	9·9	2·5	11·2	—	118
Estimated Percentage of Total Mongols for whom Birth Order was Unascertained(*b*) ..	0	7	9	18	14	22	28	41	15	24	—	—

(*a*) Obtained by multiplying row above by 1·24 (118/(118−23))
(*b*) Row above expressed as percentage of top row.

(2) Possible Interpretations of Results

If findings such as have been presented here can be freed from any type of methodological error and are repeated consistently in similar studies they are of considerable aetiological significance. McKeown and Record (1956) point out that association between incidence and parental age or birth order provides strong presumptive evidence of environmental influence, even if the action of this influence were confined to the germ cells and expressed itself through mutations in ways outlined by Penrose (1955). Similarly, an association between incidence and ordinal position points to some form of environmental influence. It would appear that any characteristic not uniformly distributed among different birth ranks and ordinal positions cannot be attributed solely to genetic transmission (although random distribution of itself is not proof of such genetic causation).

Penrose (1955) has drawn attention to the importance of parental age in differentiating causes of mutation, and he notes that the influence of the father's age is of critical significance. When there is an association between father's age and incidence (as in achondroplasia), the hypothesis of fresh gene mutation as the cause is strengthened, whereas when there is no such association (as in mongolism) other causes must be sought. He also postulates that a slight (approximately equal) increase in the mean ages of both parents would be expected in conditions originating in mutations caused by radiation.

McKeown and Record (1956) distinguish between types of environmental influence operative at different stages of development. They note that it is the early pre-natal environment that seems to determine the incidence of mongolism and anencephaly, while there is evidence to suggest that the intra-natal environment (*e.g.* relative anoxaemia) may affect the incidence of patent ductus arteriosus, and that the post-natal environment may affect the onset of infantile hypertrophic pyloric stenosis. These authors state that, unlike the uterine environment, the post-natal environment favours children in low birth ranks and at late maternal ages (due to the absence of older siblings who may convey infectious disease, and to the more favourable economic circumstances of parents). However, it may be questioned whether these generalizations apply equally to psycho-social environmental factors which may influence the development of certain psychiatric disorders.

In the light of such considerations as the foregoing, we may now proceed to examine a few of the possible interpretations that may be placed on specific findings that have emerged in the present study.

Parental Age.—In *Norton's psychiatric patients*, there was a slight increase in mean and median maternal ages (as compared with controls and general population), more marked than the apparent slight associated increase in median paternal age. The difference between the median ages of father and mother was somewhat low for these psychiatric patients, which would render mutation an unlikely hypothesis (Penrose, 1955). On the other hand, the excess among the patient group of mothers one or two years older than fathers might favour the hypothesis of maternal dominance throughout the individual's development as a possible aetiological factor.

Family Size.—The corrected mean size of family for *Heron's insane patients*, as calculated by Greenwood and Yule (1914), was 4·17 which would appear to be an unduly low figure for the children of parents in England and Wales married a considerable time before the end of the 19th century (Table VII).

Mean family sizes hitherto calculated for juvenile delinquents (Glueck and Glueck, 1950), neurotics (Norton, 1952) and schizophrenics (Wahl, 1954, 1956) all appear to be biased estimates (appreciably larger than figures that would have been obtained through samples of their mothers). Malzberg (1950) used the Greenwood–Yule correction in calculating mean family sizes on his group of mongols and obtained 2·9 as the mean size of all families (a number of which were probably incomplete) and 4·2 as the mean size for the presumably completed families of mothers aged 40 years and over.

It is known that the incidence of certain mental disorders is related to social class (*e.g.* Redlich, Hollingshead, Roberts, Robertson, Freedman, Myers 1953; Hare, 1955), and it is also well established that size of family is related to social class. It might therefore seem logical that the mean size of family for a given category of psychiatric patients should be related to the social class from which that category is predominantly derived. The mean family size of *Norton's neurotic patients* is indeed compatible with this hypothesis. However, if this hypothesis were true, it would be expected that schizophrenics derived predominantly from lower socio-economic strata than manic-depressives would also tend to come from larger families, whereas the opposite was observed in *Malzberg's two groups*. It appears, therefore, that Malzberg's patients with

dementia praecox came from unduly small families, or his manic-depressives from unduly large families, or possibly both.

Before leaving the topic of family size, it is appropriate to mention the situation of *the only child*, which should be considered in this context rather than in relation to ordinal position (*vide supra*). It has already been noted that neither of Malzberg's series show an undue representation of families with only one member, but that both Norton's psychiatric patients and his physically ill controls exhibit an unexpectedly high frequency of only children. It is possible that this may be a characteristic of the local population from which they were drawn, but it appears at least as likely that there is a raised incidence of certain types of both psychiatric and "physical" illness among only children.

Birth Order.—Greenwood and Yule (1914) found no appreciable inequality between observed and expected frequencies of the various birth ranks among *Heron's insane patients*, although admittedly the latter group were presumably heterogeneous and probably included some cases with epilepsy and mental deficiency (Malzberg, 1938).

However, though subsequent investigations have shown associations between birth order and some congenital abnormalities (as mentioned earlier), no consistent association has so far been demonstrated between birth order and the incidence of functional psychiatric disorders. Thus Malzberg (1938) obtained negative findings in *schizophrenia* (supported by Böök's findings on a much smaller series, 1953) and in *manic-depressive psychoses*. These findings appear to be corroborated by a current study of patients admitted to mental hospitals in Western Ontario (Wanklin, 1957).

In *Norton's psychiatric patients* (1952) the excess of observed over expected frequencies in the later birth ranks may be significant of itself, but appears to be partly attributable to the specific increase in observed incidence in youngest children (*vide infra*).

Ordinal Position.—Previous investigators have examined the percentage distribution of cases among the various ordinal positions, with which they have also included only children (*e.g.* Glueck and Glueck, 1950; Norton, 1952; Wahl, 1954, 1956). The only significant information that this form of distribution gives would appear to consist of possible differences in the observed percentages of eldest and youngest children (which should be approximately identical if the incidence in both is the same). Böök (1953) and Wanklin (1957) have also compared observed and expected frequencies of eldest and youngest children in small series of psychiatric patients, without obtaining statistically significant results.

However, the results of investigation of ordinal position in the four samples analysed here, by the extension of the Greenwood-Yule method that has been described, are suggestive and provocative. The most significant of these findings is an excess of observed over expected incidence in youngest children of families with four or more members, most marked in Malzberg's two series of psychotic patients. If statistical bias can be excluded as the cause of these observations (*vide supra*), we are led to speculate on what peculiarities in the situation of the youngest child might render him more vulnerable to psychiatric abnormality, and the following possibilities may be suggested:

(*i*) Advanced ages of parents at the birth of the youngest child might result either in biological handicapping (as in mongolism and achondroplasia), or in relative psycho-social handicapping.

(*ii*) Termination of the parents' marriage shortly after the birth of the child (who then remains the youngest), either through death or through the separation of the parents. Norton (1952) indeed found that a significantly higher proportion of his psychiatric patients than of his controls had lost their father before they reached the age of 10 years. However, this observation leads to the question whether, in such cases, the significant aetiological situation might be economic deprivation, loss of a normal paternal relationship, or consequent unopposed maternal dominance with accentuation of any pathological features in the mother-child relationship.

(*iii*) The youngest child might well be such because of deliberate family limitation on the part of the parents, and would thus appear to be the most likely member of the family to have been unwanted. In this event, the parents might harm the child before birth by unsuccessful attempts at abortion, or after birth by behaviour resulting from conscious or unconscious rejection. In this connexion, Sears (1957) has found (with a considerable degree of statistical confidence) that both mothers and fathers were more likely to have been displeased at the mother's becoming pregnant with the youngest child of larger families than with any other. It may also be relevant to mention here that the Royal Commission on Population (1954,

vol. VI, part I. p. 273) reported that over 17 per cent. of women in England and Wales had more than the number of children they had planned, so that the problem is one of considerable magnitude.

(*iv*) Sears (1957) has made some other interesting observations on child training according to the sex and ordinal position of the children. His findings appear to indicate that youngest children are not the coddled "babies" of the family, but that they are least often breast-fed, severely weaned, and rarely praised, and that "modesty" training is begun early.

In concluding these remarks on the possible significance of findings with regard to parental age, family size, birth order, and ordinal position, the comments of McKeown and Record (1956) seem most appropriate—namely, that the examination of such variables is of limited interest unless the significance of an observed association is explored further; that the same association may have very different explanations in different circumstances; and that, when taken with other clinical and pathological evidence, such information may suggest profitable lines of inquiry.

Summary

The investigation of parental age, family size, birth order, and ordinal position offers hope of information on the relative influences of genetic and environmental factors in the aetiology of mental disorders. Some of the relevant literature concerning these variables has been reviewed.

Certain data relating to these variables, previously recorded by two other investigators, have been subjected to further analysis. These data involve groups of psychiatric patients and physically ill controls seen in a general hospital in London and two groups of psychotic patients admitted to a mental hospital in New York State.

The Greenwood-Yule method of determining family size and expected frequencies of birth orders (from samples taken through members of the sibships) has been outlined, together with an extension of this method devised to estimate expected frequencies of different ordinal positions.

A slight, but statistically significant, increase in mean and median maternal ages was noted for Norton's group of predominantly neurotic patients, together with an apparent (though less marked) increase in median paternal age. There was also a significant excess of these patients (as compared with controls) having mothers one or two years older than their fathers.

The mean family size of Norton's psychiatric patients was approximately the same as that of his controls and of the section of the general population with which both samples were considered comparable, but the frequency of only children was unexpectedly high in both groups.

The mean family size of Malzberg's manic-depressives slightly exceeded that of his group of patients with dementia praecox and that of the general population.

Norton's psychiatric patients showed a relative excess of observed over expected incidence in the higher birth orders, but no such association with birth order was noted for the other three samples examined.

Analysis of ordinal position revealed an excess of observed over expected incidence in youngest children of families containing four or more members, most marked in Malzberg's two series of psychotic patients.

The foregoing results have been discussed, with reference to a number of possible sources of error, and some alternative explanations have been considered.

It is concluded that further investigations of such variables as parental age, family size, birth order, and ordinal position are indicated, and may well yield profitable information.

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